UTILITY OF 3D PRINTED CARDIAC MODELS IN CONGENITAL HEART DISEASE

by

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Abstract

Congenital heart disease (CHD) encompasses a heterogenous group of lesions whereby the anatomic structures and relationships of the heart have not undergone a normal development. CHD is the most common congenital malformation in newborns, occurring in about 1% of live births. Over the last 5 decades improvements in surgical and interventional catheterization techniques have resulted in an average life expectancy well into adulthood.

Three-dimensional (3D) modelling is capable of producing a physical 3D model from a digital clinical imaging data set. 3D printed cardiac models may be especially useful for the study and treatment of CHD as they can convey patient-specific information in 3D space.

This research provides insights on the scope of use of patient specific 3D printed cardiac models used in the care of patients with CHD with respect to the available current body of literature and in clinical practice. This thesis investigates 1) reported uses of 3D printed CHD models in the literature; 2) access to and use of 3D printing technology for CHD in clinical practice; 3) applications of 3D printed CHD models for interventional cardiac procedural planning and 4) applications for undergraduate medical education.

Lay Summary

Congenital heart disease (CHD) is a condition babies are born with, where the heart and/or vessels around the heart do not develop properly. Some CHDs may have complicated connections between the chambers of the heart and surrounding vessels. Three-dimensional (3D) printing is a new technology that can produce a 3D object from a digital file. 3D printing can be applied to CHD to produce patient specific heart models from a patient's clinical imaging. These models can be particularly beneficial, because they allow the user to physically manipulate the model and appreciate a patient's specific anatomy in 3D space. In this project we investigated 1) uses of 3D printed CHD models reported in the literature, 2) access and use of 3D printing technology in pediatric cardiology clinical practice, 3) use of models to simulate cardiac procedures and 4) use of 3D heart models in education for medical trainees.

Preface

All of the work was conducted at The Children's Heart Centre, The Department of Medical Imaging and The 3D Technologies Program at the Digital Lab at BC Children's Hospital (BCCH), University of British Columbia.

The project entitled "Use of Patient Specific 3D Cardiac Models to Treat Congenital Heart Disease in Clinical Practice: A Physician Survey Study" was approved by the University of British Columbia's Research Ethics Board (certificate # H19-00806). By completing the online survey, participants provided consent to participate in the research. The project entitled "Do Patient Specific 3D Heart Models Improve Outcomes for Children Undergoing Interventional Cardiac Catheterization?" was approved by the University of British Columbia's Research Ethics Board (certificate # H18-03115). All participants provided written informed consent. The project entitled "3D Printed Cardiac Models for Cardiac Medical Education" was approved by the University of British Columbia's Research Ethics Board (certificate # H19-02607). All participants provided written informed consent.

This thesis is divided into 6 chapters. Chapter 1 is a narrative literature review on congenital heart disease and three-dimensional (3D) printing.

Chapter 2 is a scoping review of the current body of literature on the uses of 3D printed cardiac models in congenital heart disease. A version of chapter 2 has been accepted for publication in Heart as Illmann CF, Ghadiry-Tavi R, Hosking MCK, Harris KC. Utility of 3D Printed Cardiac Models in Congenital Heart Disease: A Scoping Review. Illmann CF was responsible for concept formation, search strategy design, data collection, analysis and manuscript composition. Ghadiry-Tavi R contributed to data collection. Harris KC was the supervisory author on this project and was involved in concept formation. All authors contributed to manuscript edits.

Chapter 3 is a physician survey on access to and use of 3D printing technology in clinical practice. A version of chapter 3 has been published in the Canadian Journal of Cardiology Open [Illmann CF, Hosking MCK, Harris KC, Utility and access to 3D Printing in the Context of Congenital Heart Disease: An international physician survey study. CJC Open doi: https://doi.org/10.1016/j.cjco.2020.01.008]. Illmann CF conducted study design, data collection and analysis, and manuscript composition. Hosking MCK was involved with early study design. Harris KC was the supervisory author on this project and was involved in concept formation and study design. All authors contributed to manuscript edits.

Chapter 4 is a randomized, controlled pilot study on the use of 3D printed cardiac models for pre-procedural interventional cardiac catherization simulation and its effect on procedure related radiation exposure. Illmann CF conducted study design, data collection, analysis and composition. Harris KC and Hosking MCK were involved with study design. Both performed the procedures and pre-procedural simulation. Culham G was involved with study design, CT acquisition and fabrication of the 3D printed cardiac models. The 3D printed cardiac models were courtesy of the 3D Technologies Program at the Digital Lab at BCCH.

Chapter 5 is a randomized, controlled trial investigating the effect of 3D printed cardiac models in undergraduate medical education of congenital heart disease. Illmann CF conducted study design, data collection, analysis and composition. Harris KC was involved with study design. Hosking MCK was involved with study design and compilation of the teaching and testing materials. Vijayashankar S taught the congenital heart disease workshops associated with the study. Culham G was involved with study design, CT acquisition and fabrication of the 3D printed cardiac models. The 3D printed cardiac models were courtesy of the 3D Technologies Program at the Digital Lab at BCCH.

Chapter 6 is a narrative conclusion on the current and potential future uses of 3D printed cardiac models in congenital heart disease.

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List of Abbreviations

3D	Three-dimensional
3D-CM	Three-dimensional cardiac model
BCCH	British Columbia Children's Hospital
CHD	Congenital heart disease
CoA	Coarctation of the aorta
CT	Computed Tomography
d-TGA	d-Transposition of the Great Arteries
PA	Pulmonary artery
RVOT	Right ventricular outflow tract
TOF	Tetralogy of Fallot

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This research would not have been possible without the help from many intelligent people around me. I'd like to offer my sincere gratitude to the members of my supervisory committee, Dr. Kevin C. Harris, Dr. Martin C.K. Hosking, and Dr. Gordon Culham. Thank you for your mentorship and guidance in developing and carrying out this research. A special thank you to my supervisor, Dr. Kevin Harris, for supporting my goals, giving me the opportunity to design new research projects, and challenging me to think critically at every step along the way.

To the Harris Lab- Ms. Nicole Hemphill, Ms. Mimi Kuan, Ms. Cameryn Meloche, Mr. Jimmy Lopez - thank you for being great lab mates, always willing to lend a helping hand, and for making our office a welcome place to arrive every morning. And thank you, Dr. Saketh Vijayshankar for your active collaboration and endless enthusiasm.

I would like to thank all the participants and their families for taking part in the studies that made up this project. I would like to acknowledge the BC Children's Hospital Research Institute for providing funding through the BC Children's Hospital Graduate Studentship (#6553) and the Rare Disease Foundation for providing funding through a microgrant (#2913).

Special thanks are owed to the 3D Technologies Program at the Digital Lab at BC Children's Hospital for providing the 3D printed cardiac models used for this thesis.

Dedication

To my family - Mom, Dad, and Andrew- thank you for always believing in me and encouraging me to chase my dreams. And thank you for teaching me the value of hard work, dedication and perseverance.

1 Introduction

1.1 Congenital Heart Disease

Congenital heart disease (CHD) is the leading congenital malformation in newborns with a worldwide incidence of about 1% (1), and a Canadian prevalence of about 13 in 1000 children (2). CHD encompasses a heterogenous set of structural cardiac abnormalities that are present at birth and can range in complexity from simple atrial or ventricular septal defects to outflow tract obstructions and single ventricle physiologies (3–5). These structural cardiac lesions often have complex three-dimensional (3D) relationships between cardiac structures, which are patient specific. Children with moderate and complex CHD will often require one or more cardiac procedures throughout their lifetime to treat their condition. These procedures, such as open heart surgery or interventional cardiac catheterization, can correct the physiology but can result in further alterations to their 3D relationships (3). Understanding these 3D relationships is important to guide treatment, because altered cardiac anatomy results in altered cardiac physiology (6). However, these complex 3D spatial arrangements can be difficult for the user to interpret or conceptualize when represented on two-dimensional screen, as with conventional clinical imaging. A 2011 study examining participants' perception of an object's 3D orientation based on two-dimensional cross-sections, showed that participants were more likely to make errors in judgement when multiple parameters (pitch and yaw) were varied (7).

1.2 Three-Dimensional Printing

3D printing is a relatively new technology that can be used to create a physical 3D object from a digital data set (Fig. 1.1). The process of 3D modelling is multi-step and involves image acquisition, image segmentation, 3D rendering, 3D printing, followed by post processing (8). 3D printed models provide additional information to the user over a virtual 3D rendering, in that they can be physically manipulated and tactilely examined. Unlike virtual 3D renderings, 3D printed models do not require the user to interpret 3D information represented on a two-dimensional screen.

3D modelling has been used for medical applications since the late 1990s. Some applications include fabrication of patient specific anatomical models where clinical images are used as the base data sets, creation of patient-specific custom implants, and to serve as molds for prosthesis. Maxillofacial and orthopedic surgery were the first subspecialties to report use of 3D models beginning in the late 1990s and early 2000s (9). Since then, many other sub-specialties have adopted this technology including general surgery, dental, cranial surgery, otolaryngology, cardiology and cardiac surgery (9).

3D printing technology has the potential to be particularly useful when applied to pediatric cardiology due to the complex and patient-specific nature of some CHD. 3D cardiac models created based on a patients' clinical imaging data set can capture and represent patient-specific anatomical information in 3D space. Fabrication of patient specific 3D CHD models was first reported in the early 2000s (9). In 2006, Noecker et al. used digital CT data sets to construct patient specific cardiac models of 12 children, with and without congenital heart disease (10). Patient specific 3D printed CHD models have since been used in surgical planning (11–13) and education (14,15) with a few case reports in the field of pediatric cardiac catheterization (16) .

1.3 Aims and Objectives

The broad aim of this thesis is to explore how patient specific 3D cardiac models can be used for CHD management and teaching. Each body chapter of this thesis has a related specific aim.

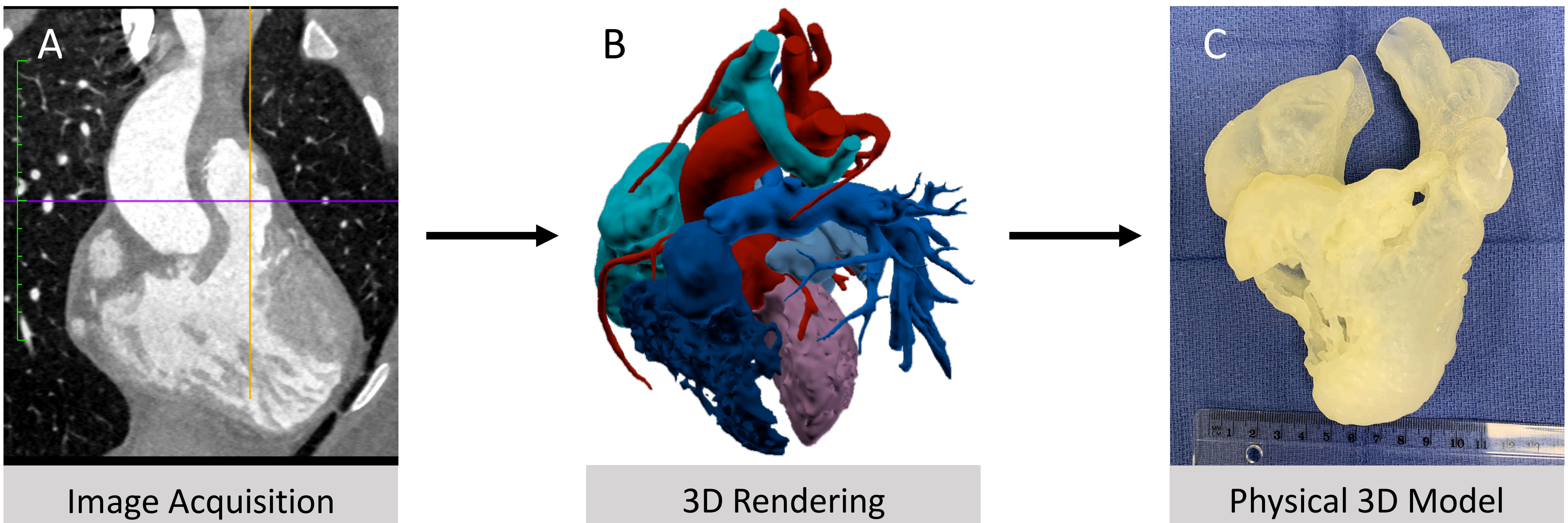
Chapter 2 is a scoping review of the literature where we sought to quantify and qualify the current body of literature reporting uses of 3D printed CHD models.

Chapter 3 is a survey of pediatric cardiologists. From this, we aimed to quantify access to 3D printing technology and determine spectrum of use of 3D printed cardiac models in clinical practice.

Chapter 4 is a randomized, controlled pilot study where patient-specific 3D cardiac models were used in pre-interventional cardiac catheterization simulating stent deployment or percutaneous pulmonary valve implantation. This study aimed to assess the effect of pre-procedural simulation on procedure related radiation exposure and procedure duration. We hypothesized that pre-procedural simulation with a 3D cardiac model would lower procedure-related radiation exposure and procedure time compared to the standard care.

Chapter 5 is a randomized, controlled study assessing the impact of 3D printed cardiac models used in a didactic workshop for undergraduate medical students. It assessed the effect of learning with 3D models on learners' knowledge acquisition and retention, measured with an objective learning assessment.

Figure 1.1 Illustrative schematic to represent the 3D modelling process.
3D modelling involves multiple steps including (A) image acquisition, manual and/or automatic segmentation to create a (B) 3D rendering, printing via a 3D printer and post-processing to achieve the final (C) physical 3D model. 3D model and rendering are courtesy of the 3D Technologies Program at the Digital Lab at BC Children's Hospital (BCCH).



2 Utility of 3D Printed Cardiac Models in Congenital Heart Disease: A Scoping Review

2.1 Introduction

Three-dimensional printing (3DP) is a new technology that allows the user to build an accurate physical 3D model (Fig. 2.1) from a digital data file. Depending on the intended use or application of the 3D model, a variety of materials can be used (17). This technology has been applied to numerous areas within the medical field, including for fabrication of patient specific anatomical models based on clinical imaging data sets, custom implants and as molds for the building of prostheses (9).

Congenital heart disease (CHD) is the most common congenital abnormality in newborns, with a worldwide prevalence of 9 in 1,000 live births (1). Some CHD lesions can have complex intracardiac spatial relationships which may be further complicated by prior surgical or interventional procedures (18). 3D printed cardiac models (3D-CMs) may aid users in fully understanding these multifaceted 3D spatial relationships, which can be difficult to conceptualize on a two-dimensional display (7). This scoping review sought to explore the applications and spectrum of use of 3D-CMs and identify knowledge gaps within the current published body of literature to guide future research.

2.2 Materials and Methods

We conducted a scoping review that targeted all published literature on the use of CHD 3D-CMs. A search strategy was constructed and tested in consultation with a research librarian to ensure adequate scope. The databases of MEDLINE, EMBASE, and Web of Science were searched from their inception until July 19, 2019. The search strategy included medical subject headings and key words separated by Boolean operators (Appendix A). Results were exported to Mendeley (Elsevier, London, UK) where duplicates were removed.

Inclusion criteria were primary research studies reporting the use of 3D-CM(s) depicting CHD lesions, cardiac models fabricated by 3DP, and human subjects. Case studies and series were included. There were no restrictions on language or year of publication. Studies published in non-English languages were translated by persons with medical knowledge. Exclusion criteria included studies where 3D-CMs were generated for proof of concept but not utilised, studies focused on bioprinting or tissue engineering technology. studies where only virtual or computational 3D-CMs were used, and non-primary research.

To determine eligibility, studies were initially screened by title and abstract followed by full-text assessment. Both steps were conducted in duplicate by two reviewers (CFI and RGT). Articles deemed ineligible by both reviewers were excluded. Disagreements were discussed between the reviewers. For disagreements that could not be resolved between the two reviewers, a third reviewer (KCH) was consulted.

Study details were recorded into a data extraction form. Details included research methodology, study aim, study population, use of 3D-CMs, main findings. Data were stored using a REDCap database hosted at BC Children's Hospital. Results were analyzed in R (R Core Team, 2018) using descriptive statistics.

2.3 Results

The search strategy initially yielded 648 results. Following assessment, 79 articles were included in the final qualitative synthesis (Fig. 2.2). A summary of included articles can be found in Appendix B and full reference list in Appendix C. Of the 79 papers included in the final review, the largest proportion (30%) originated from the USA (Table 2.1). The majority of studies (n=52, 66%) were case series or reports that were descriptive in nature without a priori hypotheses. Only 12 articles (15%) reported use of a control group. The first article was published in 2006 with the number of publications increasing yearly until 2017, which had the greatest number of publications. Publications reported use of computed tomography (68%), magnetic resonance imaging (37%) and 3D echocardiography (6%) as the base imaging dataset for the 3D-CMs. 13 articles reported use of multiple imaging modalities. 4 studies did not report which imaging modality was used to construct the 3D-CMs.

We identified 3 broad areas in which 3D-CMs are being used: procedural planning, procedural simulation and education (Table 2.2). 3D-CMs were most commonly used for procedural planning (72%). More specific categories were surgical planning, interventional cardiology (IC) planning, surgical simulation, IC simulation, medical education and patient and family education. It was possible for an article to fall into one or more of these areas (Fig. 2.3).

2.3.1 Surgical Planning

37 articles reported the use of 3D-CMs for surgical planning. Of these, 31 were case reports [Appendix C. 1-20] or small case series [Appendix C. 21-31]. The remaining 6 articles were primary research studies with pre-defined aims and outcome measures. These articles generally sought to quantify the benefit of 3D-CMs in terms of change in surgical procedure [Appendix C. 32,33], operative time [Appendix C. 34,35], and accuracy of 3D-CM [Appendix C. 36,37].

The case reports and series document use of 3D-CMs in pre-surgical planning of corrective, palliative and transplant procedures for a range of complex CHD lesions (Appendix B). These studies largely concluded that 3D-CMs were useful to define the spatial complexity and relationships between intracardiac structures [Appendix C. 2,3,6,7,10,11,19,22,31] and relationships between the heart and adjacent structures [Appendix C. 13]. 3D-CMs were used to determine feasibility of surgical management [Appendix C. 14,19,24,31] and to plan optimal approach [Appendix C. 1,3-5,13,16,25,26] including pre-determining optimal protheses design, conduit or patch size and site [Appendix C.15,18,26,27]. In some cases, authors report the 3D-CMs were sterilized, brought into the operating room, and used for intraoperative orientation [Appendix C. 9,10,25,28-30]. In addition, some case reports speculate that use of 3D-CMs reduced operative time [Appendix C. 20,27]. A shortcoming of 3D-CMs is that they cannot depict fine intracardiac structures such as the cardiac valves and chordae [Appendix C. 21,26].

There were 5 prospective interventional studies and one retrospective chart review that evaluated the use of 3D-CMs for surgical planning. Valverde et al. and Xu et al. investigated the effect of 3D-CMs on procedural plan. They found that the procedural plan in 19 of 40 patients and 2 of 15 patients, respectively, changed after evaluation of patient-specific 3D-CMs [Appendix C. 32,33]. Zhao et al. and Ryan et al. investigated the impact of pre-surgical planning with 3D-CMs on operative time. Both groups reported lower operative time in cases where 3D-CMs were used in planning. However, neither reached statistical significance [Appendix C. 34,35]. Ma et al. and Ngan et al. evaluated the accuracy of 3D-CMs compared to intraoperative findings in conjunction with surgical planning. They concluded that 3D-CMs accurately represented the patients' anatomy [Appendix C. 36,37].

2.3.2 Surgical Simulation

8 articles report use of 3D-CMs for surgical simulation. 4 utilised surgical simulation for pre-surgical planning [Appendix C. 5,6,16,23], 1 reported surgical simulation for surgical training [Appendix C. 38], and the remaining 3 articles demonstrate proof of concept that 3D-CMs can be used for surgical simulations [Appendix C. 39-41]. Case reports identified that 3D-CMs and simulation helped with understanding of the 3D anatomy and could be used to simulate intraoperative view [Appendix C. 5,23,40].

There are limitations on quality of 3D-CMs for use in surgical simulation. Yoo et al.'s study of 81 surgeons and trainees identified that cardiac valves were poorly represented on the 3D-CMs and the print material differed in consistency and elasticity from human myocardium [Appendix C. 38]. This concern was echoed by Ilina et al. who reported a method to produce silicone cardiac valve models with 3DP molds. 6 cardiac surgeons used these models to simulate complete atrioventricular canal defect repair. The silicone cut and held sutures well but was more rigid than human tissue [Appendix C. 41].

2.3.3 Interventional Cardiology Planning and Simulation

25 articles reported the use of 3D-CMs for IC planning [Appendix C. 14,17,22,24,25,42-61]. The majority of these (n=16) utilised simulation of the intended procedure as part of the pre-procedural planning [Appendix C.42-47,49-52,57-61]. Of the 25 articles that reported use of 3D-CMs for IC planning there were 17 case reports [Appendix C. 14,17,43,44,46,48-54,57-59,61,62], 6 case series [Appendix C. 22,24,25,42,45,47], 1 retrospective comparison [Appendix C. 55], and 1 small controlled trial [Appendix C. 44]. These descriptive studies largely report that planning or simulating an IC procedure with a 3D-CMs is feasible.

Within the setting of IC, 3D-CMs were used to plan device closure [Appendix C. 25,42,45,50,51,53,57,59-62], stent placement [Appendix C. 43,44,48,49,58], percutaneous valve replacement [Appendix C. 47,55], cardiac ablation [Appendix C. 54], develop novel procedures [Appendix C. 47,52] and in clinical decision making for optimal procedural management [Appendix C. 14,17,22,24,25]. Multiple articles report the benefit of using 3D-CMs over conventional IC planning techniques was that optimal device size and position could be tested on the 3D-CM and accurately predicted prior to procedure [Appendix C. 43,44,46,57-59,61].

Matsubara et al.'s controlled trial of 11 patients, investigated the impact of pre-procedural simulation of patent ductus arteriosus device closure using a hollow 3D-CM. During the simulation, device size and type were selected. The authors report lower fluoroscopic and procedure time in the study group. However, their study was under-powered and statistical analysis was not conducted [Appendix C. 60]. Schievano et al. retrospectively fabricated 3D-CMs for 12 patients who were previously referred for pulmonary valve implantation (PPVI). With the 3D-CMs, 2 cardiologists, who were blinded to the known clinical outcome, were better able to predict PPVI success than with MR images alone [Appendix C. 55].

2.3.4 Medical Education

3D-CMs have been used for CHD education in multiple populations of health care professionals including pre-medical students [Appendix C. 63], medical students [Appendix C. 2,63-65], paediatric residents [Appendix C. 66-68], nurses [Appendix C. 69-71], surgical trainees [Appendix C. 38], cardiac surgeons [Appendix C. 38], and ancillary care providers [Appendix C. 71]. The 3D-CMs have been incorporated into educational initiatives in various ways including didactic teaching sessions [Appendix C. 64-67], simulation-based training programs [Appendix C. 38,63,68] and educational initiatives at point-of-care [Appendix C. 2,71].

3D-CMs have been well received by learners [Appendix C. 2,67] and shown to aid their learning experience [Appendix C. 70]. However, studies have reported heterogenous conclusions with regard to learners' knowledge acquisition after teaching with 3D-CMs [Appendix C. 2,63,65,67,68]. Multiple studies have reported a positive correlation between lesion complexity and learners' knowledge. These findings suggest that 3D-CMs may be most beneficial for use in education of complex over simple CHD lesions [Appendix C. 64,66,71].

Endpoints to evaluate the efficacy of 3D-CMs in medical education are not standardized between studies. Various outcome measures have been used to determine the impact of 3D-CMs on learning have been used. Some of these outcome measures include subjective self-reported knowledge [Appendix C. 63-66,68,71], objective knowledge assessments [Appendix C. 65-67], learner-reported satisfaction [Appendix C. 67] and qualitative feedback from users [Appendix C. 2,38,69].

2.3.5 Patient and Family Education

The use of 3D-CMs for patient and family education has been reported in trials specifically designed to investigate their impact on learners' knowledge [Appendix C. 69,72,73] and as adjuncts within case reports where 3D-CMs were originally manufactured for procedural planning [Appendix C. 2,20,74]. The case reports anecdotally report that the 3D-CMs can aid patients and families to understand their underlying CHD anatomy and the need for procedural intervention [Appendix C. 2,20,74]. This is supported by qualitative studies that reported patient-specific 3D-CMs were easier for parents to understand than clinical images or illustrated 2D diagrams. They propose that 3D-CMs could help with engagement during consultation [Appendix C. 69,73].

There have been varied reports on impact of 3D-CMs on learners' objective knowledge. A randomized controlled study of 97 parents of patients with CHD found no difference in parental objective knowledge between those who had been counselled with or without 3D-CMs [Appendix C. 73]. An uncontrolled study of 20 adolescent CHD patients found that patients' objective knowledge significantly increased following a clinic visit with 3D-CMs. Since this study did not utilise a control group, the effect of the clinic visit on patient knowledge cannot be separated from the effect of the 3D-CM [Appendix C. 72].

2.4 Discussion

2.4.1 Quality of Evidence

3D-CMs have been used in a spectrum of applications for CHD including procedural planning, procedural simulation and education. Most of the published literature of 3DP and CHD for pre-procedural planning are case reports, small case series and uncontrolled interventions that report anecdotal findings and lack rigorous statistical analysis. These small descriptive reports have successfully demonstrated the feasibility of using 3D-CMs to reproduce complex CHD anatomy for procedural planning and simulation. However, these reports may have led researchers to prematurely conclude that there are clinical benefits from use of 3D-CMs in procedural planning over standard of care. Some of these cited benefits include reduced procedure time [Appendix C. 27,34,60], cardiopulmonary bypass time [Appendix C. 27,34] and radiation exposure [Appendix C. 60] despite a lack of statistically significant findings or control group. This reflects the broader context that most current studies lack the statistical power to determine the magnitude or direction of impact that 3D-CMs have on procedures and care of patients with CHD. Without a control group or comparing to population based normative data, such a conclusion is speculative. There is clearly opportunity for future research to build on this early work by conducting prospective controlled trials to delineate the role of this technology. One of our aims is to identify these gaps in the literature and provide a roadmap for future research (Table 2.2).

Most of the experimental studies assessing 3D-CMs are within the areas of surgical planning and medical education [Appendix C. 32-38,63-65,67,68,70]. Within these areas there remains room for improvement in terms of defining meaningful outcome measures. Several educational studies used subjective feedback and self-assessed knowledge as primary outcome measures [Appendix C. 2,63,64,66,68,73]. These are open to response bias whereby the participants may be inclined to self-report higher knowledge after participating in a learning opportunity with 3D-CMs (19). Some surgical planning studies used case-crossover study design to assess if procedural planning using 3D-CMs could change clinical management decisions [Appendix C. 32,33]. But, with lack of a true control group there is no way to fully clarify if this surrogate measure of change in planned procedure is clinically beneficial for the patient. Thus, there is a need for future controlled studies with pre-defined objective outcome measures.

2.4.2 3D Printed Cardiac Models Convey Complex 3D Spatial Information

The key benefit of using physical 3D-CMs over standard images displayed on a 2D screen may be the ability to represent intricate and complex relationships between cardiac structures in 3D space. In the studies where 3D-CMs were used for procedural planning, numerous case reports and series describe the proceduralists' improved ability to appreciate spatial complexity and relationships with use of a patient specific 3D-CMs [Appendix C. 2,3,6,7,10,11,13,19,22,31]. When used in education, medical students and residents reported higher knowledge aquisition when learning with a 3D-CM representing complex over simple CHD lesions [Appendix C. 64,66]. Retrospective and prospective investigations using 3D-CMs in procedural planning compared to conventional imaging modalities using 2D alone, impact [Appendix C. 33,55] and may potentially improve clinical decision making.

3D-CMs may not be appropriate for all current applications. Participants from a surgical simulation training course with 3D-CMs found that cardiac valves were poorly represented, and print material did not resemble human myocardial consistency [Appendix C. 38]. In CHD lesions where valve attachments and chordae greatly affect management decisions, 3D-CMs should be used in conjunction with 3D and 4D echocardiography to encompass a comprehensive understanding of valve structure, function and its impact on adjacent structures (20) [Appendix C. 38]. Dynamic structures (e.g. valve apparatus that change position throughout the cardiac cycle) and those that are very fine (e.g. chordae tendineae, fossa ovalis) are not well suited to 3D modeling due to suboptimal characterization of these structures on image data sets used to generate the models (21).

2.4.3 Future Areas of Utilization

A potential future area of utilisation of 3D-CMs is for education of allied health professionals. To date, there are reports of 3D-CMs for education in pre-medical students [Appendix C. 63], medical students [Appendix C. 2,63-35], paediatric residents [Appendix C. 66-68], combined paediatric/ emergency residents [Appendix C. 66], nurses [Appendix C. 69-71], surgical trainees [Appendix C. 38], and cardiac surgeons [Appendix C. 38]. Bhatla et al. anecdotally reported that a 3D-CM helped an echocardiographer to guide the probe and obtain optimal views during the patient's echocardiogram [Appendix C. 19]. Conceivably, 3D-CMs could be incorporated into educational programming for echocardiographers or other allied health professionals who work with patients with CHD. Furthermore, IC simulation with 3D-CMs has only been used in conjunction with IC planning. There is potential for IC simulation on 3D-CMs to be used in medical training. It has been shown that 3D-CMs used for surgical simulation have been an effective tool in surgical education [Appendix C. 38].

We speculate that there is variation in users' ability to conceptualize complex 3D spatial relations and intuitively extrapolate anatomic relationships from 2D image data sets. The true benefit of 3D-CMs may be related to the users' innate ability to understand 3D spatial relationships and interact with technology. Future research could investigate which groups benefit the most from 3D-CMs compared to conventional 2D imaging modalities and other novel virtual 3D technologies, such as virtual reality (22), 3D imaging overlay (23), or holography (24).

2.4.4 Breadth of Review

To our knowledge, this is the largest scoping review to investigate the utility of 3D-CMs for use in CHD to date. The search was conducted in a systematic fashion, however there is a possibility that some articles may have been missed. There are a few systematic reviews in this area with narrower scopes of focus on surgical planning (25,26), image segmentation (27) or limitations on year of publication and language (28). In agreement with our scoping review, others have concluded that the majority of evidence consists of case series and case reports and there is a need for future studies to provide evidence-based information to guide the use of 3D-CMs in clinical practice (26,28). The 2018 systematic review by Lau et al. was limited to 28 papers published in English between 2007-2017 (28). Our contemporary review is more current and had no restrictions on year or language of publication. There remains a paucity of high-quality controlled studies to provide a suitable evidence base to guide clinicians and educators on how best to make use of this technology.

2.5 Conclusions

3DP is a new technology that has been widely used and integrated into the care of children with CHD over the past 15 years. The primary reported areas of utilisation of 3D-CMs include procedural planning, procedural simulation, and education with the most reports in the realm of procedural planning. The majority of the published reports are experiential in nature where the authors reported the use of 3D-CMs but do not define a priori research questions or outcome measures. Reports to date lack sufficient evidence to determine whether there is a real benefit of 3D-CMs over standard care or other innovative health technologies, such as virtual reality (22) or 3D imaging overlay (23). There is need for future evaluation to establish best use of this technology, which should be approached with a robust scientific process. Our study has identified gaps in the literature and addressed priority areas for future research.

Figure 2.1 3D Printed Cardiac Model
3D-CM depicting right sided heart structures of a patient with previously repaired tetralogy of Fallot. 3D-CM was used for procedural planning and simulation of percutaneous pulmonary valve implantation.
RA= right atrium; RV= right ventricle; RVOT= right ventricular outflow tract

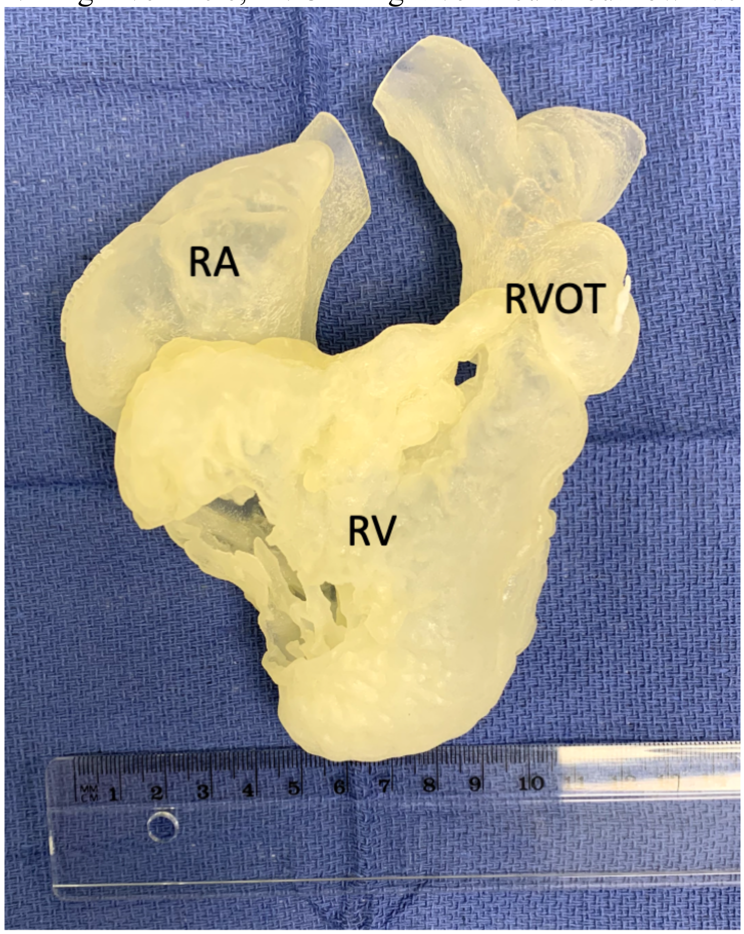


Figure 2.2 PRISMA Flow Diagram
Flow diagram of articles considered for study inclusion.

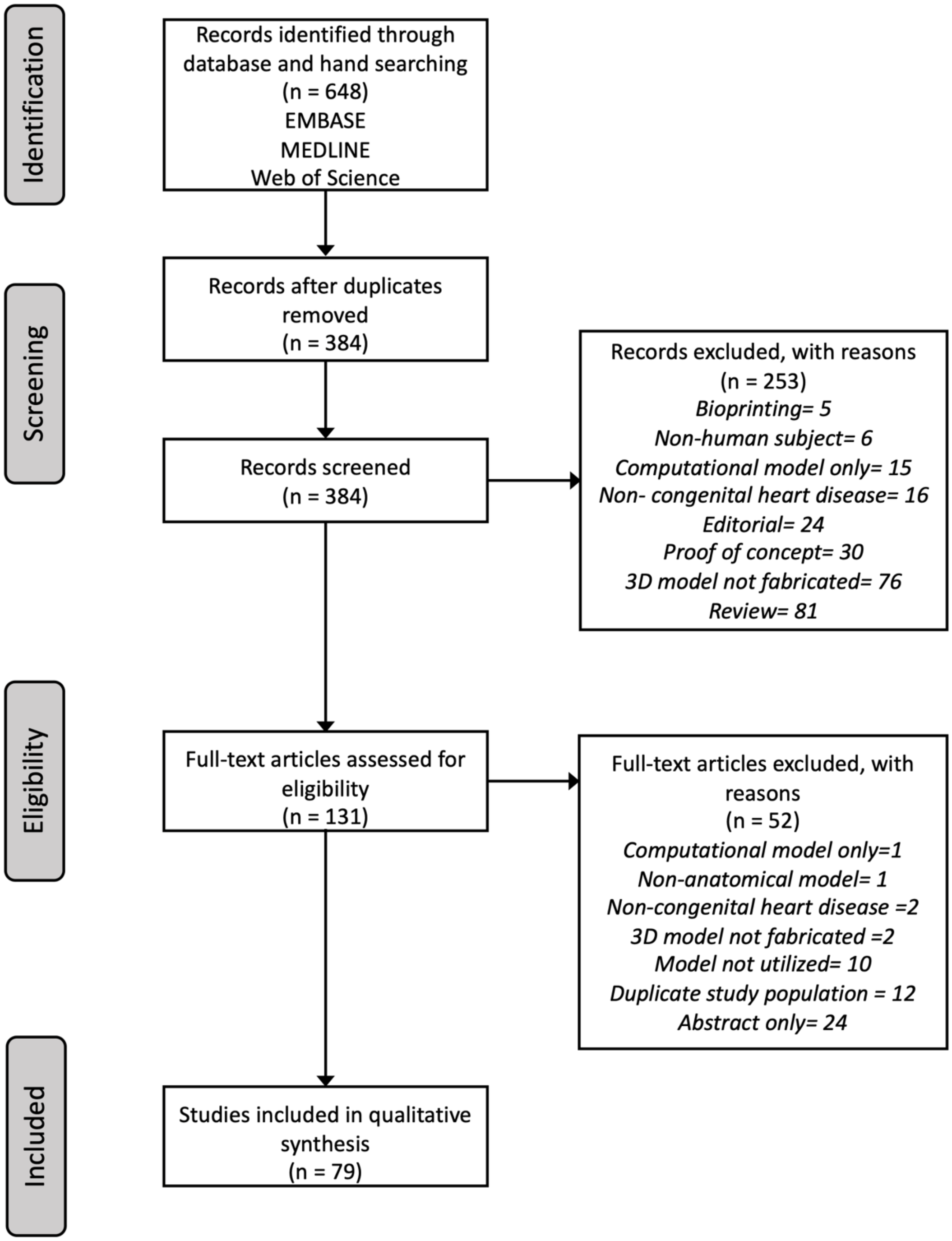


Figure 2.3 Area of Utilisation Upset Plot

Studies fell into the areas of surgical planning, interventional cardiology planning, surgical simulation, interventional cardiology simulation, medical education, and patient and family education. It was possible for an article to fall into one or more of these areas. Figure 2.3 illustrates the number of articles per area of utilisation and the intersection of areas of utilisation per article.

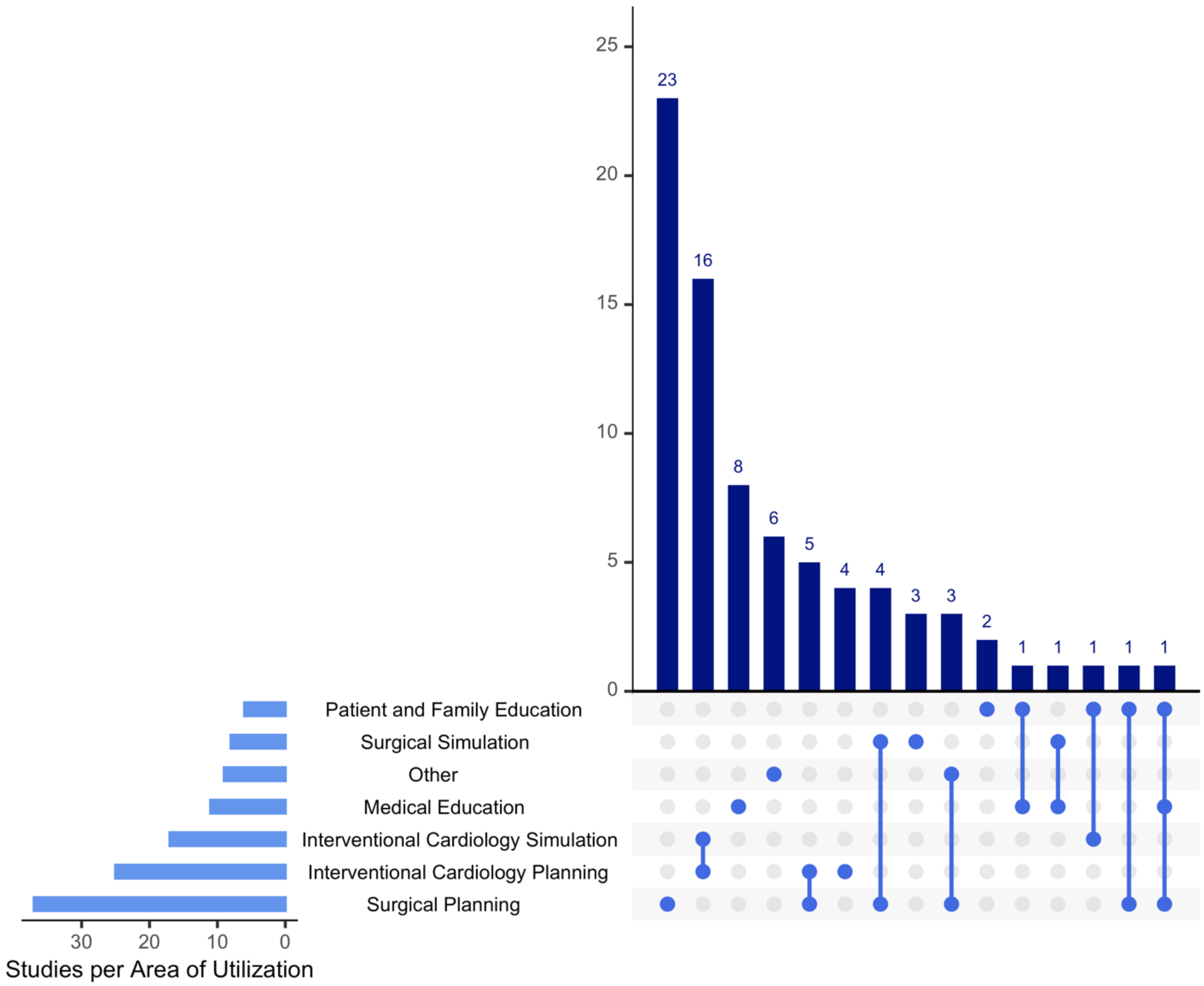


Table 2.1 Number of articles by country of origin.
*Reference numbers refer to reference list of included studies in Appendix C

Country of Origin	**Number of Articles**	**References***
Argentina	1	[Appendix C. 10]
Belgium	1	[Appendix C. 20]
Canada	2	[Appendix C. 37,38]
China	11	[Appendix C. 32,34,36,42,45,46,59,61,62,65]
France	1	[Appendix C. 3]
Germany	6	[Appendix C. 9,26,28-31]
Hungary	1	[Appendix C. 21]
India	4	[Appendix C. 1,14,27,76]
Ireland	2	[Appendix C. 18,24]
Italy	2	[Appendix C. 43,74]
Japan	6	[Appendix C. 5,23,40,58,50,60]
Poland	2	[Appendix C. 17,54]
Slovakia	1	[Appendix C. 22]
Spain	4	[Appendix C. 8,12,15,48]
United Arab Emirates	2	[Appendix C. 6,56]
United Kingdom	7	[Appendix C. 2,55,57,69,70,72,73]
United States of America	24	[Appendix C. 4,7,11,13,16,19,25,35,39,44,47,49,51-53,63,64,66-68,71,77-79]
International Collaboration	2	[Appendix C. 33,41]

Table 2.2 Knowledge gaps and areas for future research in 3D printing for CHD.

*Number of studies in each area does not equal total number of studies included in the final qualitative synthesis because some studies covered multiple areas of utilization; **Reference numbers refer to reference list of included studies in Appendix C.

<table>
<tr><th colspan="2">Spectrum of Use*</th><th>What do we know?**</th><th>Gaps and Limitations**</th></tr>
<tr><td rowspan="2">Plan</td><td>Surgical Planning
(n=37)</td><td>• 3D models are useful to appreciate the spatial complexity between intracardiac structures [Appendix C. 2,3,6,7,10,11,19,22,31]
• Use of 3D models can influence a change in procedural plan (surgical vs. conservative management) [Appendix C. 33]</td><td rowspan="2">• Unclear if procedural planning with 3D models can improve clinically meaningful outcomes such as reduction in operative time [Appendix C. 34-35], readmission [Appendix C. 35], procedure related radiation exposure [Appendix C. 60], or mortality rate [Appendix C. 35]
• Models cannot accurately depict valves and chordae [Appendix C. 21,26]
• Need for appropriately powered prospective research</td></tr>
<tr><td>Interventional Cardiology Planning
(n=25)</td><td>• 3D models can be used in clinical decision making for optimal procedural management [Appendix C. 14,17,22,24,25,55]</td></tr>
<tr><td rowspan="2">Practice</td><td>Surgical Simulation
(n=8)</td><td>• Surgical simulation has been used in conjunction with surgical planning [Appendix C. 2 5,6,16,23] and surgical training [Appendix C. 38]
• Models aided understanding of 3D anatomy and can be used to simulate intraoperative view [Appendix C. 5,6,23]</td><td>• Print material consistency differs from human myocardium [Appendix C. 38]
• Cardiac valves were poorly represented [Appendix C. 38]</td></tr>
<tr><td>Interventional Cardiology Simulation
(n=17)</td><td>• Optimal device size and position could be tested on the 3D model and accurately predicted prior to procedure [Appendix C. 43,44,46,57-59,61]</td><td>• Potential to use interventional cardiology simulation in educational initiatives for interventional cardiology trainees</td></tr>
<tr><td rowspan="2">Educate</td><td>Medical Education
(n= 11)</td><td>• 3D models may be most helpful for learners to understand complex CHD lesions over simple lesions [Appendix C. 64,66,71]</td><td rowspan="2">• Unclear if 3D models improved learners' objective knowledge [Appendix C. 2,63,65,67,68] or knowledge retention over time
• 3D models may be useful in education of allied health professionals such as echocardiographers</td></tr>
<tr><td>Patient and Family Education
(n=6)</td><td>• 3D models can aid patients and families in understanding their underlying CHD anatomy and the need for procedural intervention [Appendix C. 2,20,74]</td></tr>
</table>

3 Utility and access to 3D Printing in the Context of Congenital Heart Disease: An international physician survey study

3.1 Introduction

The medical application of three-dimensional (3D) printing technology is a rapidly developing field for children with congenital heart disease (CHD) (27). The usage of 3D printed cardiac models have been reported in a wide variety of settings including patient and family education (29,30), medical education (14,31–38), pre-procedural planning (12,39,40) and procedural simulation (16,41). CHD is heterogenous, patient specific and varied with respect to 3D spatial relationship between structures. The models can convey intricate and nuanced information about the 3D spatial relationship between cardiac structures that may not be well appreciated in conventional 2D imaging modalities. This enhanced 3D spatial information can have implications on procedural decision making and in turn, patient outcomes.

We sought to determine 1) the current spectrum of use of 3D printed cardiac models in the congenital heart disease 2) the access to 3D printing technology for pediatric cardiologists.

3.2 Materials and Methods

3.21 Study design and population

This was a cross-sectional survey targeting pediatric cardiologists who treat patients with CHD. Questionnaires were disseminated and responses were collected between May and September 2019. A voluntary response sampling methodology was used. Members of the Canadian Pediatric Cardiology Association (CPCA) and Congenital Cardiac Interventional Study Consortium (CCISC) were eligible to participate. To recruit participants, an email including study rationale, invitation to participate and a link to the online questionnaire was sent to the members of CPCA and CCISC. Questionnaires were distributed using Research Electronic Data Capture (REDCap) software hosted at BC Children's Hospital. REDCap is a secure, web-based software platform designed to support data capture for research studies, providing 1) an intuitive interface for validated data capture; 2) audit trails for tracking data manipulation and export procedures; 3) automated export procedures for seamless data downloads to common statistical packages; and 4) procedures for data integration and interoperability with external sources (42,43). This study was approved by the BC Children's Hospital research ethics board. By completing the questionnaire, respondents acknowledged they were giving consent to participate in the research. All responses were received anonymously.

3.2.2 Questionnaire

The questionnaire contained 25-items that enquired about physician's access to 3D printing technology, experience using 3D printed cardiac models and opinions on best uses of patient specific 3D printed cardiac models. Prior to dissemination to CPCA and CCISC, the questionnaire was distributed to pediatric cardiologists and pediatric cardiology research staff at BC Children's Hospital to test content and branching logic. Results were analyzed using descriptive statistics, Fisher's exact test and odds ratio.

3.3 Results

3.3.1 Demographics

71 pediatric cardiologists responded to the questionnaire. 47 responses were collected from CPCA members and 24 responses from CCISC members. This represents a response rate of 57% and 10% for CPCA and CCISC, respectively. Respondents were located internationally, in

5 of the 7 continents (Figure 3.1). The gross majority (93%) practiced in North America, specifically in Canada or the United States of America (USA).

3.3.2 Pediatric cardiologists think 3D models are beneficial for children with CHD

85% (60/71) agreed or strongly agreed that patient specific 3D cardiac models are or can be a beneficial tool in treating patients with congenital heart disease (Figure 3.2). Of those that believed 3D models are beneficial tool, the leading perceived benefits of the 3D models were that they facilitated communication with colleagues (80.0%, 48/60) or with patients and their families (72%, 43/60) (Figure 3.3). 3% of (2/71) respondents disagreed or strongly disagreed that 3D models were a beneficial tool. Respondents who disagreed were able to provide justification in an open text box. One respondent justified that "3D models often cannot accurately depict valve attachments which can be an important consideration in the clinical management of a CHD patient."

3.3.3 Access to 3D printing is presently limited

49% (35/71) of respondents reported that they had access to 3D printing technology at their institution. Access to 3D printing technology was not evenly distributed between geographic location of respondents (Figure 3.4). There was a significant difference in access to 3D printing technology based on location of respondent ($p=0.004$). Of those who responded, pediatric cardiologists from the USA were 5.5 times more likely (95% CI, 1.6 to 19.2) to have access to 3D printing technology compared to Canadian pediatric cardiologists.

The primary reasons for respondents lacking access to 3D printing technology were financial barriers (50.0%, 18/36) and preference for standard 3D imaging modalities (33%, 12/36), such as 3D echocardiography (Figure 3.5). A minority of respondents without access to 3D printing, believed that the technology is not important (6%, 2/36) or that there was lack of interest at their institution (19%, 7/36). 22% (8/36) of respondents faced other reasons for lack of access to 3D printing technology. Half of these respondents (4/8) elaborated that they practiced in non-surgical centers and that 3D printing is not important in non-surgical centers and 25.0% (2/8) responded that their institution was in the process of starting a 3D printing program.

Of respondents with access to the technology, 77% (27/35) report that they have utilized the technology in the treatment of patients with CHD. Most respondents who are utilizing 3D models (96%, 26/27) report the primary use is in the conduct of clinical care as opposed to in the context of research protocols (4%, 1/27). 82% (22/27) of respondents utilizing 3D printing technology began printing within the last 4 years. The annual volume of 3D printed models produced varied by institution. The median reported volume was 5 models per year (IQR 3, 10).

3.3.4 Reported Uses

In clinical practice, the primary reported use of 3D printed cardiac models was for procedural planning. Almost all (96%, 26/27) respondents who are utilizing 3D printing technology have used models for surgical planning and approximately half (52%, 14/27) have reported using the models for interventional cardiology planning. Less than one third of respondents used 3D printed cardiac models for educational purposes and less than one quarter of respondents used models for surgical simulation (Figure 3.6).

3.3.5 Reported lesions

The most common lesion for which 3D models were utilized was double outlet right ventricle (DORV) (70%, 19/27) followed by single ventricle anatomy (48%, 13/27) (Figure 3.7).

These were also the lesions which pediatric cardiologists found the model most helpful, compared to standard of care. 90% (17/19) of respondents who had used models for DORV found the 3D models to be helpful. Use of 3D models for other lesions is more sporadic. Compared to standard of care, most respondents who have used 3D printed models (78%, 21/27) reported that the models were most helpful because they improved communication with colleagues (Figure 3.8). 44% (12/27) of respondents reported that the 3D models were helpful in that they reduced procedure or procedure planning time.

3.4 Discussion

3.4.1 Access to 3D printing is limited

Among physicians who do have access to 3D printing technology, the vast majority reported using the technology in the treatment of children with CHD. Of pediatric cardiologists without access to the technology, only a small minority claim the reason for lack of access is because they do not believe the technology is important. These finding indicate that there is acceptance of the technology amongst pediatric cardiologists and suggest that the limited access is more related to barriers to access than acceptance of technology. The primary barrier to access 3D printing technology was financial. The financial cost of 3D printing is related to the cost of 3D printing machinery and associated operating costs, including disposable equipment, maintenance and hiring skilled personnel. The capital cost of a 3D printer and associated segmentation software is variable and related to the quality of the printer and software. Printers can range in cost from a few thousand dollars for entry level printers to several hundred thousand Canadian dollars for high fidelity printers that can print in a range of materials, range of colours and have axial resolution up to 10-15 microns. The cost of segmentation software is also highly varied from free for open access software to Canadian $17,500 /year for a professional license (Materialise Mimics Innovation Suite) (44). The segmentation process of converting clinical images to STL files, that can be read by the 3D printer, is labour intensive and time-consuming that involves expertise from skilled personnel. These skilled personnel are to understand segmentation and anatomy, as well as managing a 3D printer. Since medical 3D printing is an emerging field the availability of these personal is limited and costly but may be eased as more are trained over time. A 2016 systematic review of segmentation methods used for 3D printing cardiovascular systems found that a majority of published studies utilized manual or semi-automatic segmentation methods, over fully automatic segmentation methods (27). As seen with other electronic technologies, as 3D printing technology develops over time, the price of the 3D printers is expected to decrease (45). The cost of segmentation may decrease with development of fully automatic segmentation software (27), and in conjunction with lower printer costs may lead to greater access to 3D printing technology in the field of pediatric cardiology.

3.4.2 Access to 3D printing technology varied by geographical location

Respondents from the USA were significantly more likely to have access to 3D printing technology over their Canadian counterparts. Discrepancy in access to the technology may be related to funding structures in each of the countries. We speculate that in Canada access is limited to centres where grant and/or donor funding can be secured to support the development of such a program. In the United States' market-based health care system, a 3D printing program may offer an institution a competitive advantage over others that do nott offer 3D printing. Thus, funding for a 3D printing program may be more likely to come from hospital or institutional administration (46). A similar trend in early adoption of health care technology between Canada and the United States was seen when magnetic resonance imaging (MRI) was introduced to clinical care in the 1980s. In the late 1980s, the United States had nearly 8 times more MRI units per capita than Canada (47). The combination of capital equipment and skilled human resources needed to initiate an MRI facility is similar to a 3D printing program (48).

3.4.3 In clinical practice, the primary use of 3D cardiac models is for procedural planning of complex CHD

Our study found that in practice, the most common use for 3D models of CHD lesions is for procedural planning, specifically for surgical planning. Furthermore, 11% of respondents without access to 3D printing technology felt the technology was only necessary in surgical centers. This indicates that amongst pediatric cardiologist there may be a perception that the most beneficial use of 3D models is in surgical planning, rather than uses in education, communication or simulation. Models are most commonly constructed for DORV. DORV is a complex CHD that encompasses a wide spectrum of anatomical arrangements. Classification is based on the 3D spatial relationship between the ventricular septal defect and the great arteries (49,50). These patient specific factors are critical when determining the optimal therapeutic approach in children with DORV. There are multiple surgical approaches to treat DORV and surgical decision making for primary or staged repair is highly influenced by patient's subclassification and 3D anatomic arrangement (51). The high rate of use of models for DORV may indicate that pediatric cardiologists find 3D models helpful to fully define the intricate spatial relationship between the VSD and great arteries. This technology helps facilitate communication of these critical relationships with cardiac surgeons.

3.4.4 3D models may be under-utilized in the context of medical education

3D printing technology has the potential to be utilized for multiple medical educational initiatives. It has been reported that 3D printed cardiac models successfully facilitate teaching of CHD lesions including, pulmonic stenosis, atrial septal defect, coarctation of the aorta, d-transposition of the great arteries, hypoplastic left heart syndrome (14), Tetralogy of Fallot (14,35), ventricular septal defects (37), and vascular rings and slings (34), for medical students and residents. Other potential uses of 3D printed CHD models include use for distributed medical education in rural locations and to preserve libraries of cardiac specimens (52). However, in our survey we found that less than 30% of respondents indicate they were using 3D models for medical education purposes implying that 3D printing technology may be under-utilized in education.

3.4.5 3D models facilitate communication with colleagues

We found that most of the respondents who think 3D models are beneficial, perceive the benefit to be from facilitating communication with colleagues. This was also the greatest reported benefit associated with 3D models compared to standard care, of respondents who have utilized 3D printed cardiac models in the care of children with CHD. These results echo findings from Olivieri et al. that investigated the impact of using patient specific 3D cardiac models during hand off following congenital cardiac surgery (53). In their study, health care providers rated using 3D models as more effective than standard verbal hand off. These findings indicate that the main benefit of 3D models may be in improving communication between healthcare providers. However, quantifying how well models improve communication between healthcare providers compared to standard care is subjective and challenging to measure.

This study is limited by the voluntary response sampling methodology. Since we used a convenience sampling method, there is potential for a sampling bias. Only pediatric cardiologists registered with CPCA or CCISC were invited to participate and therefore not all pediatric cardiologist who treat children with CHD were included. There is also potential for response bias, where recipients of the survey with a vested interest in 3D printing would be more likely respond. However, we received about equal numbers of responses from participants with and without access to the technology.

3.5 Conclusions

3D printing is a new technology that has been readily adopted by cardiologists who treat children with CHD. The majority of pediatric cardiologists surveyed feel that patient specific 3D printed models are a beneficial tool in the treatment of children that can be used to facilitate communication with colleagues and aid in surgical planning. However, access to 3D printing is limited and varied by geographic location. Respondents from the United States were significantly more likely than their Canadian counterparts to have access to the technology. In clinical practice, 3D models are primarily used for procedural planning for CHD lesions with complex 3D spatial relationships.

Figure 3.1 Map of Respondent Locations

For illustration purposes, the quantity of respondents relative to the respondents' broad geographic location were mapped (ArcMapTM, v. 10.6; Esri Inc., CA). ArcGIS® and ArcMapTM are the intellectual property of Esri and are used herein under license. Copyright © Esri. All rights reserved. For more information about Esri® software, please visit www.esri.com.

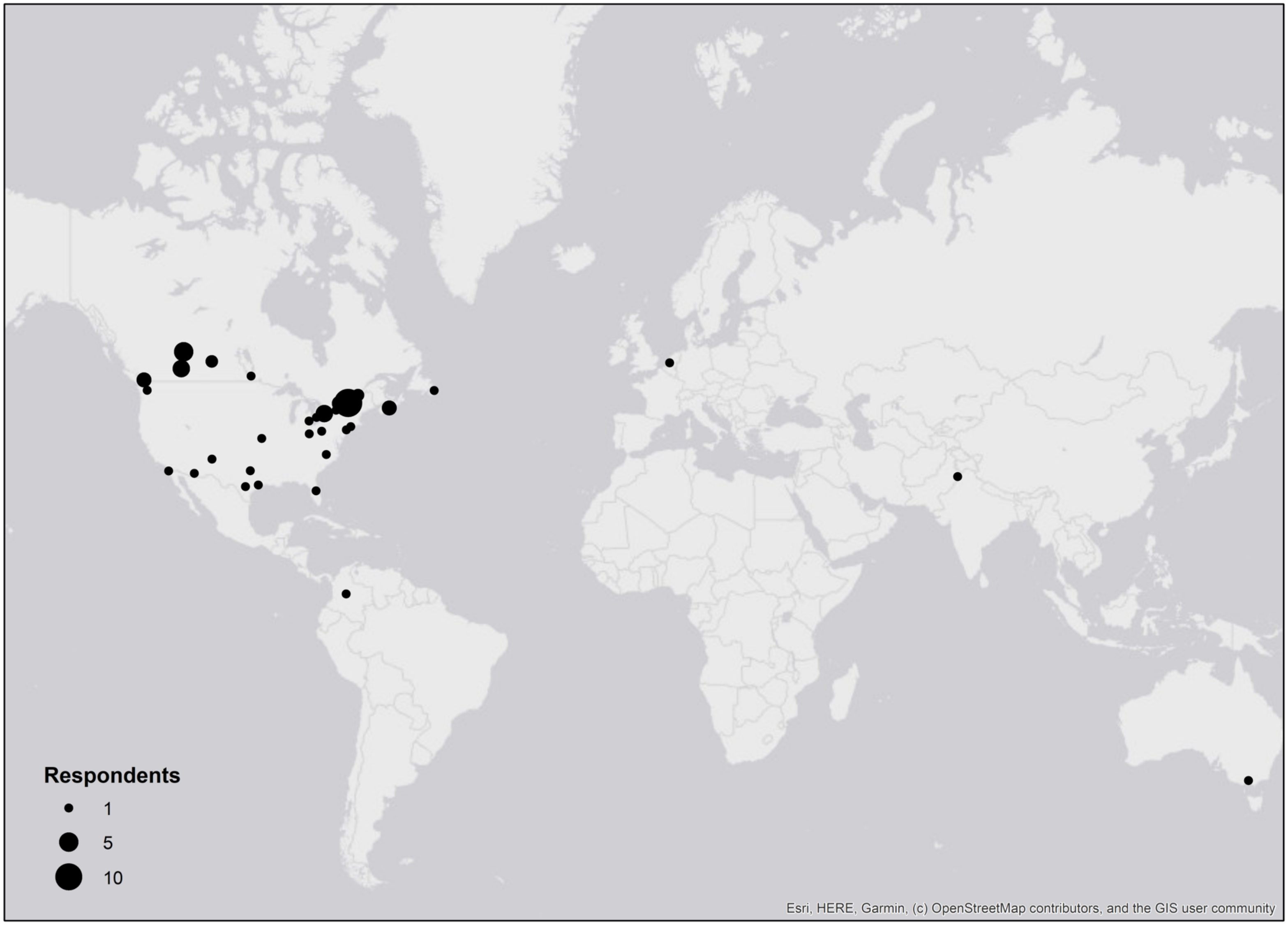


Figure 3.2 Respondents' opinion of benefit of 3D printed models.
Response to the statement "Patient specific 3D cardiac models are or can be a beneficial tool in treating children with congenital heart disease."

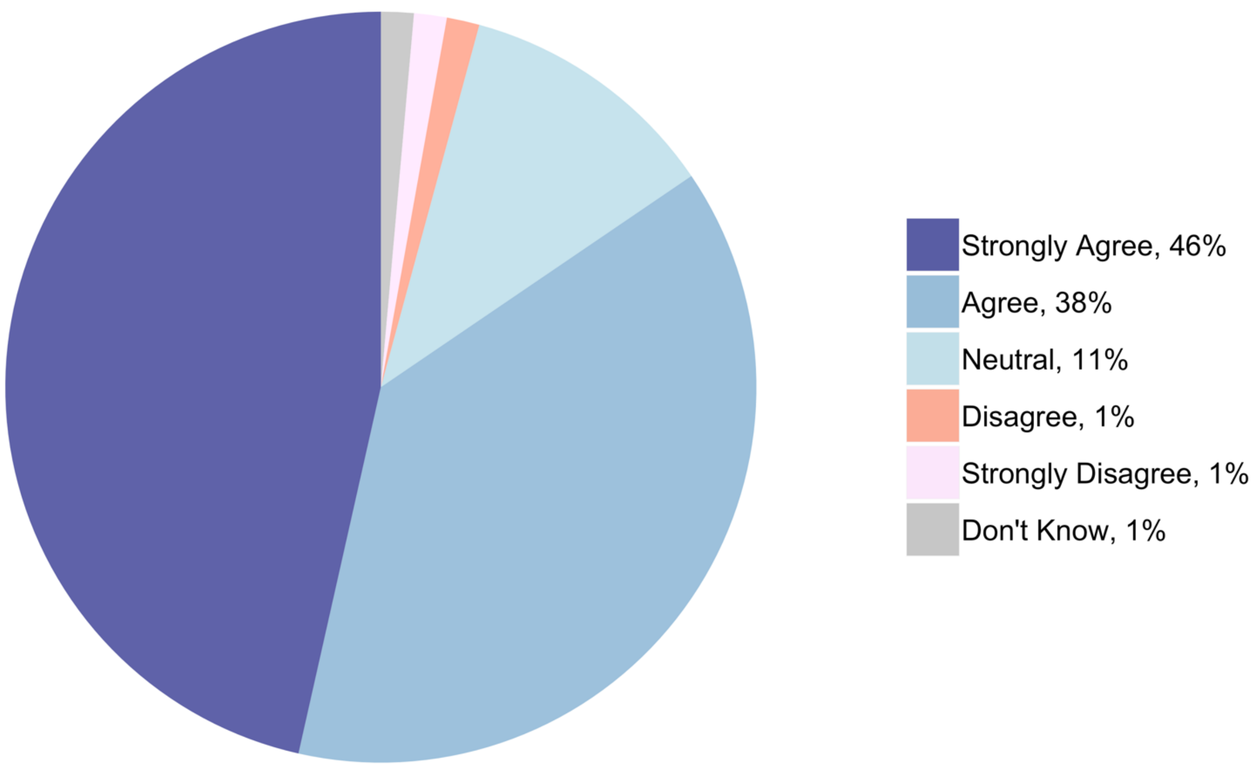


Figure 3.3 Perceived benefits of 3D printed cardiac models.
Respondents who agreed or strongly agreed that 3D models were beneficial tool in treating children with CHD were surveyed on their opinion of the perceived benefit of 3D models.

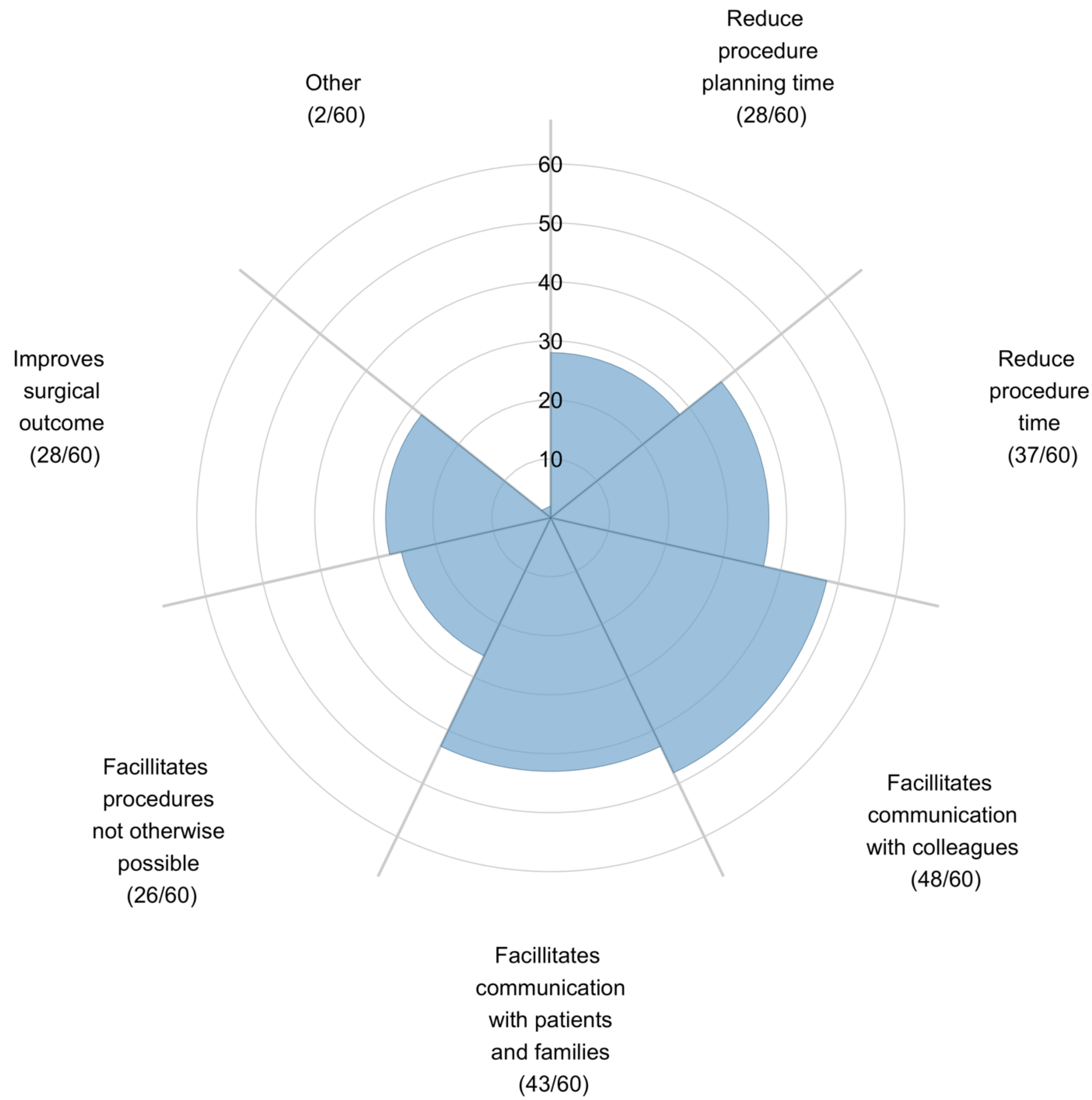


Figure 3.4 Access to 3D printing technology by country.

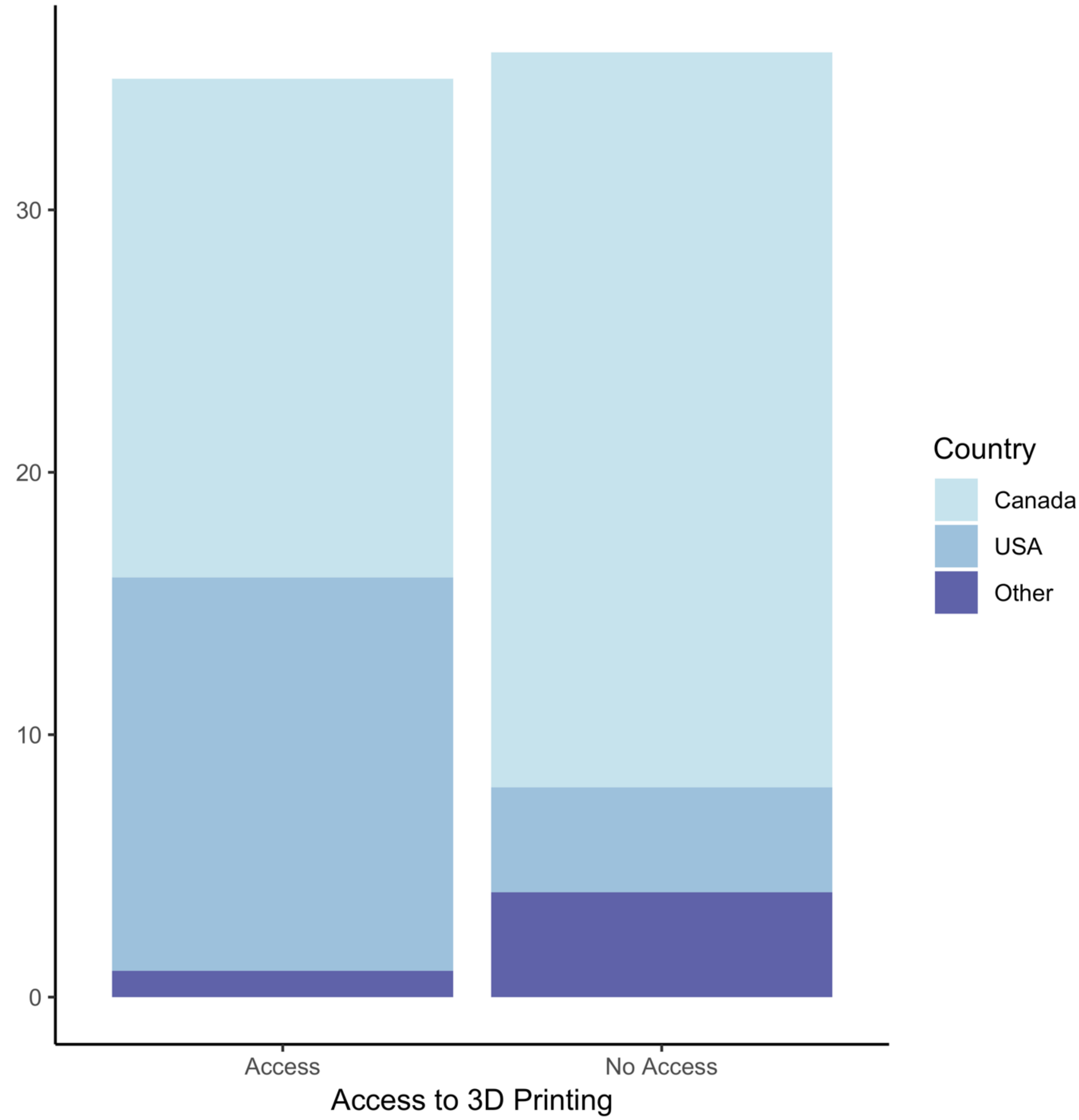


Figure 3.5 Barriers to access of 3D printing technology
Respondents without access to 3D printing technology were surveyed on the reason for lack of access.

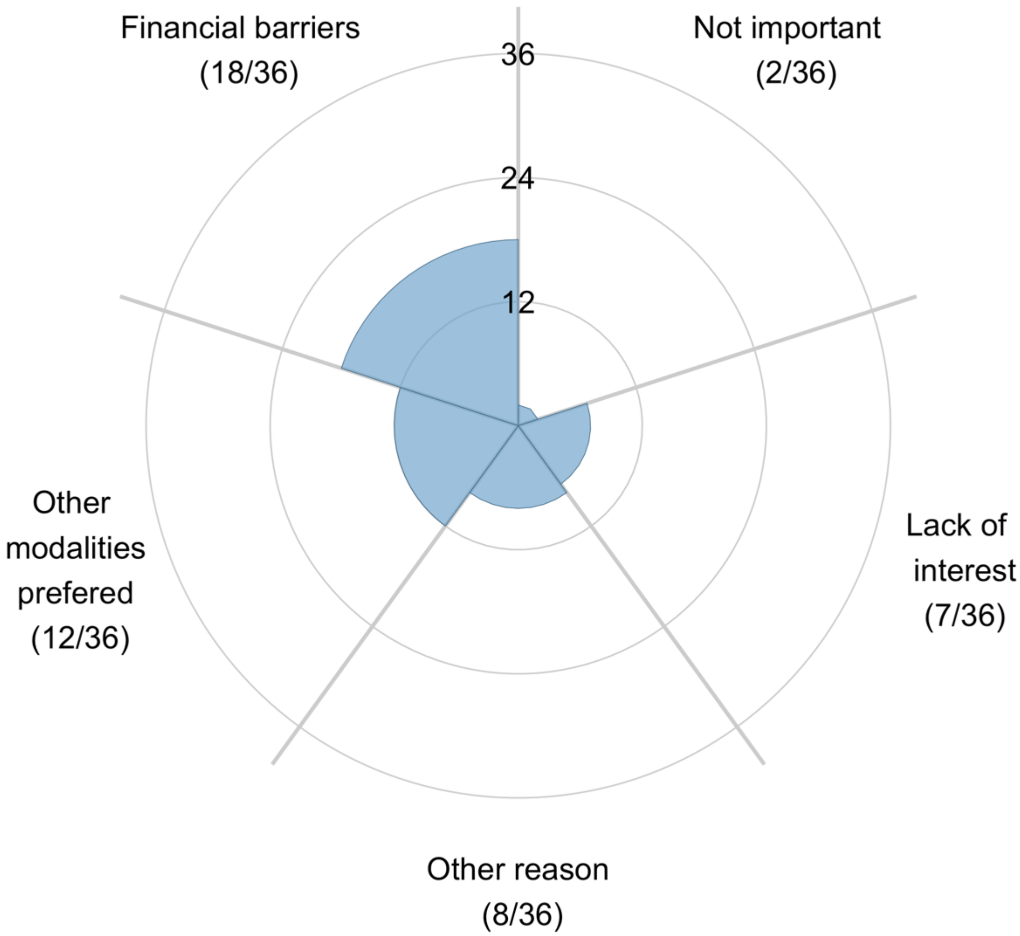


Figure 3.6 Uses of 3D printed cardiac models in clinical practice
Respondents who have access to and utilize 3D printing were surveyed on how they use the technology in clinical practice.

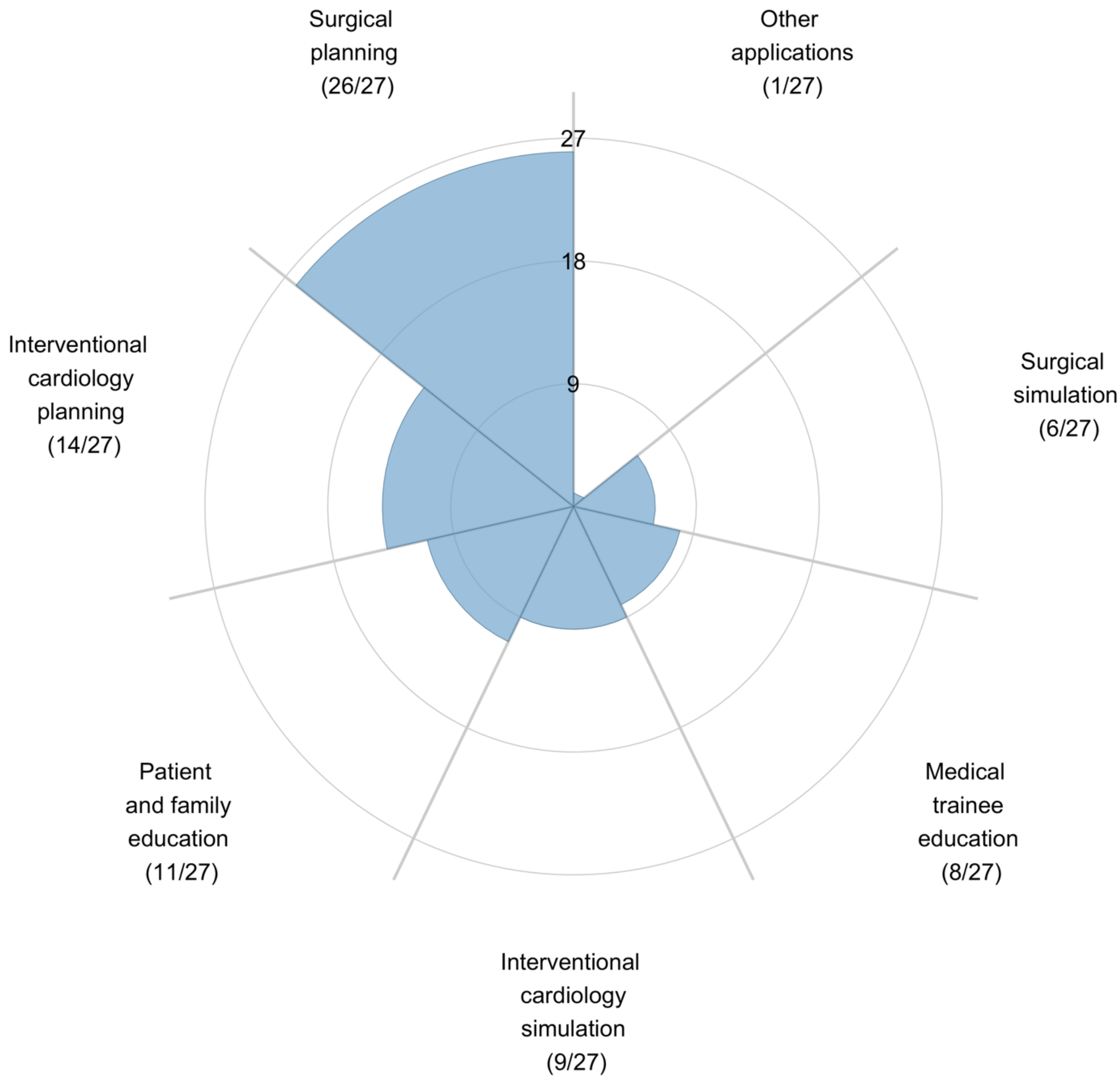


Figure 3.7 Cardiac lesions modelled with 3D printing in clinical practice
Cardiac lesions modeled with 3D printing. Dark bars indicate lesions that respondents have used a 3D model to represent. Light bars indicate the lesions that respondents found 3D models were most helpful.

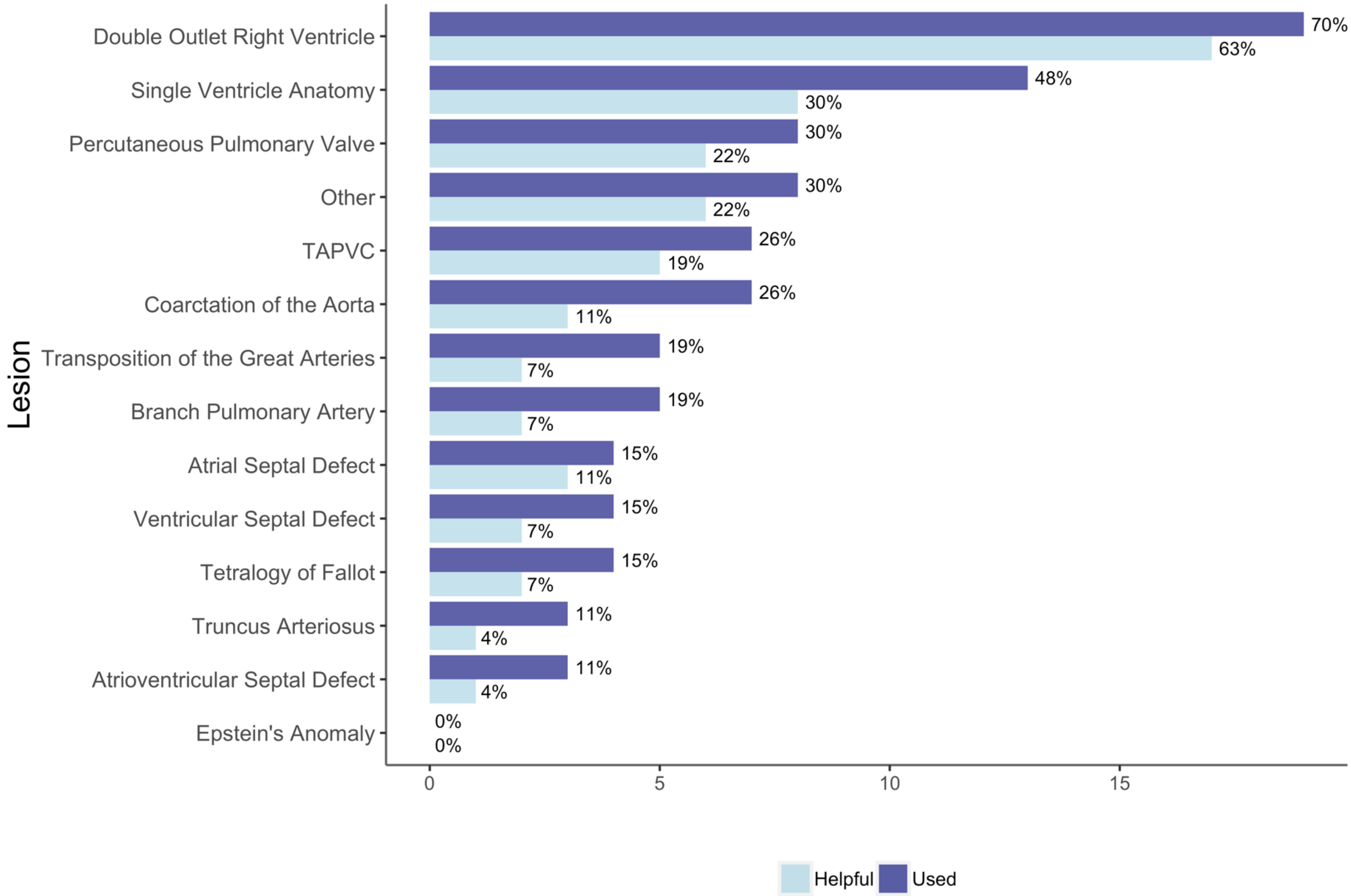
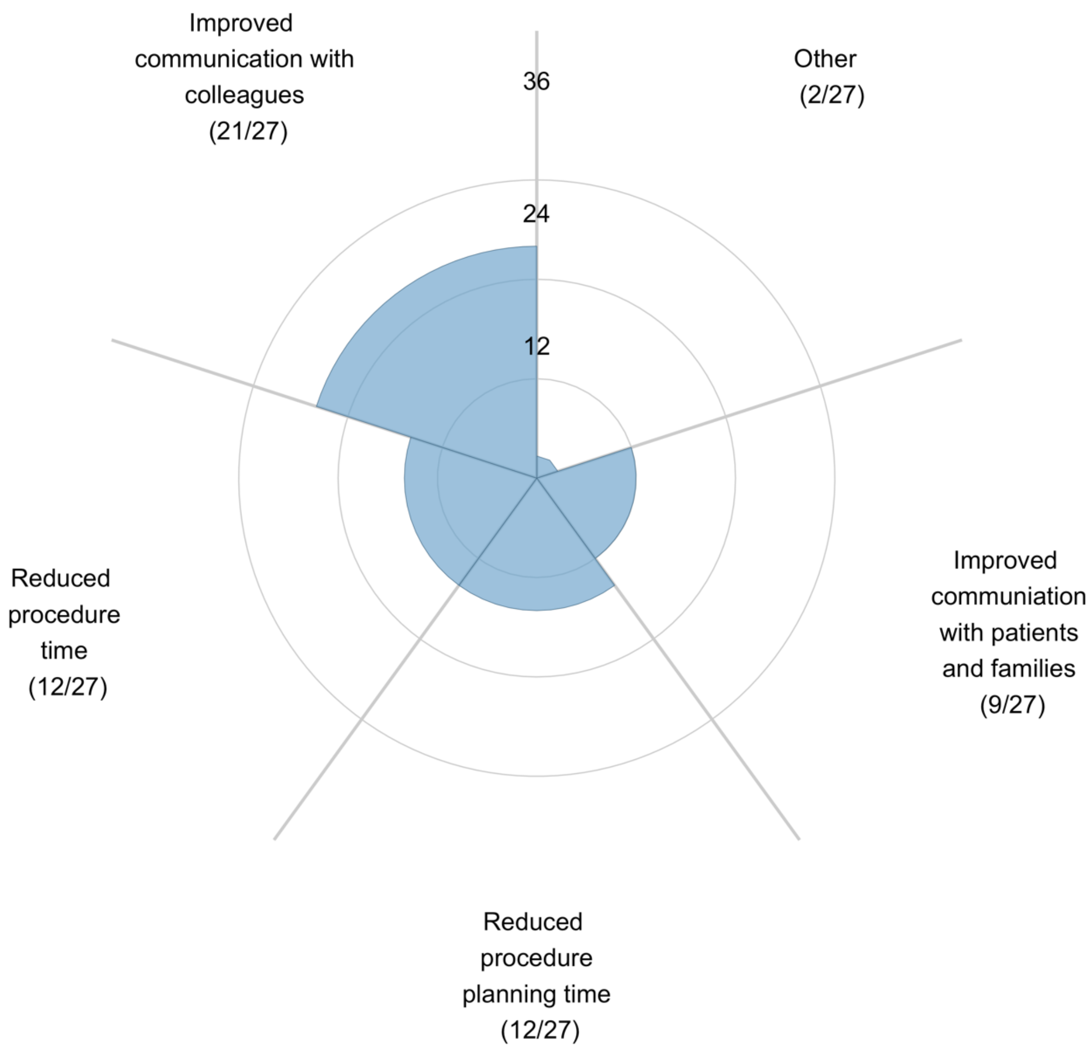


Figure 3.8 Benefits of 3D printed cardiac models

Respondents who use 3D printing technology were surveyed on how the 3D printed cardiac models are beneficial as a tool in the treatment of CHD compared to standard of care



4 Impact of Pre-Procedural Interventional Cardiac Catheterization Simulation on Radiation Exposure: A Randomized, Controlled Pilot Study

4.1 Introduction

Compared to surgery, interventional cardiac catheterization is less invasive, has shorter recovery time, and is an accepted option to treat some forms of congenital heart disease (CHD) (54,55). However, interventional cardiac catheterization introduces other risks to patients, namely procedure related radiation exposure (56,57). Radiation exposure is a known carcinogen and increased radiation exposure is directly correlated with an increased lifetime risk of cancer (58). This is a concern for pediatric cardiology patients, who are more sensitive to radiation and exposed to significant cumulative radiation doses throughout their lifetimes (59).

3D printed models can be particularly useful for the study and treatment of CHD, because the models can depict the heterogenous, patient-specific anatomic variation and nuanced 3D spatial information that might be otherwise difficult to appreciate from images represented on two dimensional screens. Previous work has shown that 3D modeling from clinical cardiac imaging data sets can produce accurate anatomical representations (10,16). These 3D models have previously been used for communication with patients and families (15), medical education (14,35), surgical planning (40) and in small case series or reports focussed on interventional cardiology planning or simulation (16,60,61).

In this pilot study we aimed to: 1) determine the feasibility of a randomized controlled trial using patient specific 3D cardiac models for pre-procedural simulation of interventional cardiac catheterization and stent placement 2) evaluate the impact of pre-procedural simulation on radiation exposure; 3) assess the impact of pre-procedural simulation on procedure time.

4.2 Materials and Methods

4.2.1 Inclusion Criteria

Patients with coarctation of the aorta (CoA), right ventricular outflow tract obstruction (RVOT) or branch pulmonary artery (PA) stenosis requiring percutaneous stent implantation or percutaneous pulmonary valve implantation (PPVI) at BC Children's Hospital (BCCH) in Vancouver, Canada were considered for this study. Participants must have previously undergone or have a cardiac computed tomography (CT) scan planned and be between 0-20 years of age (Fig. 4.1).

4.2.2 Randomization

Participants were randomized in a 1:1 ratio between 3D model and standard of care groups using stratified block randomization, with a block size of 4. Stratification was based on condition (CoA, RVOT, PA, PPVI), to ensure a similar distribution of conditions between treatment arms. With a block size of 4, there were 6 possible permutations (1: AABB, 2: ABAB, 3: ABBA, 4: BABA, 5: BAAB, 6: BBAA). Where, A represents allocation to the standard of care group and B represents allocation to the 3D printing group. The randomization block sequence was created using a random number generator within MS Excel (Microsoft, Redmond, USA) with equal probability of selecting integers 1 through 6 (62). Block assignment was blinded to the investigators and personnel conducting informed consent.

4.2.3 3D Modelling

3D models were produced based on the patient's most recent cardiac computed tomography (CT) scan. CT data sets were loaded into Mimics software (Materialise, Belgium) where the blood pool was segmented. Segmentation was performed by a skilled engineer in consultation with a cardiac radiologist and pediatric cardiologist. Segmentation was done initially by an automatic thresholding algorithm based on Hounsfield units of radiodensity, followed by manual clean-up. Following segmentation, a virtual 3D model was created and saved as a stereolithography (STL) file. The STL files were edited and optimized using 3-matic software (Materialise, Belgium). To create a hollow model, a virtual hollow shell was created around the 3D blood pool. Further editing to crop and select a region of interest, and to remove any free-floating pieces was conducted. The finalized STL file was processed on an Objet 260 printer (Stratasys, Eden Prairie, MN) to create the physical 3D model. The model was constructed out of a translucent, flexible, rubber-like material, Tango+ (Stratasys, Eden Prairie, MN) with SUP705 support material (Stratasys, Eden Prairie, MN) to support overhangs and complex geometries during the 3D printing process. The physical 3D model was post-processed to remove the support material using a 1% NaOH immersion, water jet wash and manual detailing. Post-processing was done in an iterative fashion until the desired finish was achieved and all support material was removed.

4.2.4 Model Validation

Following production and prior to simulation, the 3D model was scanned by CT. Perimeter measurements at analogous locations on the 3D model CT data set and patient's anatomical CT data set were made. CT data sets were analyzed using Horos™ software (Horos Project, Annapolis, MD USA). A fixed threshold was selected midway between the highest and lowest radiodensities (63). Measurements were compared using Pearson's correlation coefficient and Bland-Altman plot. Statistical analysis was performed in R (R Core Team, Vienna, Austria).

4.2.5 Procedural Simulation

Simulation of the intended interventional cardiac catheterization procedure, for participants randomized to the 3D model group, was performed on the patient specific 3D model prior to the participant's procedure. Simulation was performed by the treating interventional cardiologist in the same catheterization laboratory where the patient's procedure was performed (Fig. 4.2). During the simulation, the 3D model was place on the operating table in anatomic orientation of supine position, optimal fluoroscopic angle projections were determined, and the intended device was deployed under fluoroscopic guidance. The optimal fluoroscopic angle, device size, fit and stability were assessed after deployment (Fig. 4.3), recorded and were used during the participant's procedure. Participants randomized to standard care group proceeded directly to procedure without simulation.

4.2.6 Interventional Cardiac Catheterization Procedure

Procedures were performed by two experienced interventional pediatric cardiologists (KCH and MCKH). For participants in the 3D printing group, the fluoroscopic projection angles determined during procedural simulation were used during the procedure. Stent type and size determined during pre-procedural simulation were used. In addition, the 3D model was available in the cardiac catheterization laboratory and used for anatomical orientation during the procedure. Participants in the standard care group underwent interventional cardiac catherization according to current best practices. In both treatment arms, procedure time (measured in minutes), defined as time between sheath insertion and removal, was recorded. Fluoroscopy time (measured in minutes) and dose area product ([DAP] measured in μGym^2) were automatically captured and recorded by a dosimeter in the catheterization laboratory. DAP index to body weight was calculated by dividing DAP by the participant's weight in kilograms (kg).

4.3 Results

4.3.1 Recruitment

Participants were recruited between February 2019 and March 2020, at time of their scheduled cardiac CT, pre-catheterization clinic visit at BCCH, or over the phone if there was no planned appointment at BCCH. Recruitment was paused prematurely due to the international COVID-19 pandemic, and subsequent research curtailment at BCCH that came into effect on March 16th, 2020. Ten participants consented to participate in our study. 100% of patients consented to participate. Following review of clinical imaging and prior to randomization, the treatment plan changed for 4 of 10 participants to conservative or surgical management (Fig. 4.1). Six participants went on to randomization, 3 were allocated to the standard care group and 3 to the 3D model group (Table 4.1).

4.3.2 Model Validation

Due to the malleable nature of the 3D models, it was impractical to directly measure the 3D model. Direct measurement of the models with calipers proved to be highly variable and imprecise because of model compression leading us to opt to measure the models indirectly by CT. Models proved to be radio-opaque when imaged with CT (Fig. 2, Panel E-G). There was excellent correlation between 3D model and patient's medical images (Pearson's $r = 0.999$) (Fig. 4.4, Panel A). The mean difference in measurement between analogous anatomic structures on 3D model and patient's anatomy was 0.087 ± 0.045 cm. All measurements were within the limits of agreement set at 1.96 SD (Fig. 4.4, Panel B).

4.3.3 *Procedure Related Radiation Exposure*

The average DAP indexed to body weight for the 3D model group was 59.2 $\mu gy^2/kg$ compared to 127.7 $\mu gy^2/kg$ in the standard care group. The average fluoroscopy time for the 3D model group was 18.6 min compared to 28.3 min in the standard care group. These data suggest that 3D model group had lower procedure related radiation exposure in comparison to the standard care group (Table 4.1). However, the sample size of the pilot study precludes definitive statistical analysis.

4.3.4 *Simulation Stent Selection*

In two of the three patients allocated to the 3D model group, the stent size and/or type used for the patient's procedure was changed following simulation of stent placement on the 3D model (Table 4.2). For patient 2 a pre-mounted Genesis 3910 Trans-Hepatic biliary stent selected and used in the simulation on the 3D model. Following the simulation, the stability of the stent was assessed and deemed to be suboptimal. During the patient's procedure a stent with a larger diameter (Genesis XD 2910 mounted on a 14mm balloon) was selected and used. A 3.9cm CP bare metal stent mounted on a 20 mm balloon was used during the procedural simulation for Patient 7. After assessment of the 3D model with the stent in place, it was deemed that a larger stent size should be used during the procedure and a 4.5cm CP covered stent on a 22 mm balloon was used.

4.4 Discussion

Cumulative radiation exposure is an important concern for patients with CHD (59). These patients are exposed to greater levels of ionizing radiation from diagnostic imaging and interventional procedures compared to their healthy peers. Recent studies have found that compared to health controls, patients with CHD are more than twice as likely to develop cancer in their lifetime (64,65). Mandalenakis et al. found that CHD patients born in the early 1990s compared to the 1970s or 1980s had an even higher risk of developing cancer in their lifetime (HR=3.37) (64). Given the increased life-expectancy for patients with CHD in recent decades, cancer risk is a vital health concern for this population (55,66). Pre-procedural simulation of interventional cardiac catheterization using patient specific 3D models represents an innovative and promising strategy to reduce procedure-related radiation exposure for patients undergoing interventional cardiac catheterization.

To our knowledge our study is the first randomized controlled trial to test the effectiveness of pre-procedural simulation using 3D model on radiation exposure. There has only been one other controlled trial investigating the effect of pre-interventional cardiac catheterization procedural simulation on radiation exposure. Matsubara et al.'s 2019 non-randomized controlled trial used 3D printed cardiac models to simulate patent ductus arteriosus device closure. They reported 7 patients in the 3D model group and 4 patients in the control group. Similar to our study, Matsubara's group found procedure time and fluoroscopy time were lower in 3D model group compared to control. Given their small sample size, statistical analyses were not conducted (67).

Our data suggests there is a reduction in procedure-related radiation exposure when simulation is performed. This study combined with Matsubara et al.'s findings and lack of other statistically significant findings in the literature (see Chapter 2) highlights the need for further investigation on the effect of pre-procedural simulation with 3D models on radiation exposure. Future studies should be appropriately powered, randomized, and controlled to assess the true magnitude and significance of pre-procedural simulation of interventional cardiac catheterization on radiation exposure.

We selected CoA, RVOT and PA lesions for this study for the following reasons. Firstly, all of the selected lesions required pre-procedural cardiac imaging for procedure planning, using cardiac CT at our institution. This imaging data set could be used to create the patient specific 3D model, in participants allocated to the 3D model arm of the study. All lesions were treated similarly with stents (68,69). It is worthwhile to note that image acquisition by cardiac CT exposed patients to radiation (65) and is part of standard of care procedure planning at BCCH for these patients. Participants in this study were not exposed to any additional radiation over standard care. A limitation related to this study is that CT data may not show the aorta or great vessels in their maximum dimension because image acquisition collected at one timepoint throughout the cardiac cycle and is not usually synchronized with cardiac systole at our institution.

4.5 Conclusions

This single centre pilot study has demonstrated that cardiac CT data sets can be used to produce accurate 3D cardiac models. This pilot study implies that it is likely feasible to conduct a randomized, controlled trial for pre-interventional cardiac catheterization simulation to guide optimal stent selection and placement. Our data suggest that procedure related radiation exposure is lower when pre-procedural simulation with a patient-specific 3D model is conducted compared to standard care. However, the small sample size prevented statistical analyses. This trial also demonstrated that pre-procedural simulation influenced clinical decision making for the procedure (stent type, size and length). There is a need for future large, appropriately powered studies to assess the full benefit of pre-procedural simulation of interventional cardiac catherizations with 3D printed cardiac models. Data from this pilot study has been used to plan a pan-Canadian multi-centre randomized controlled trial and funding application to the Canadian Institutes of Health Research.

Figure 4.1 Study design.

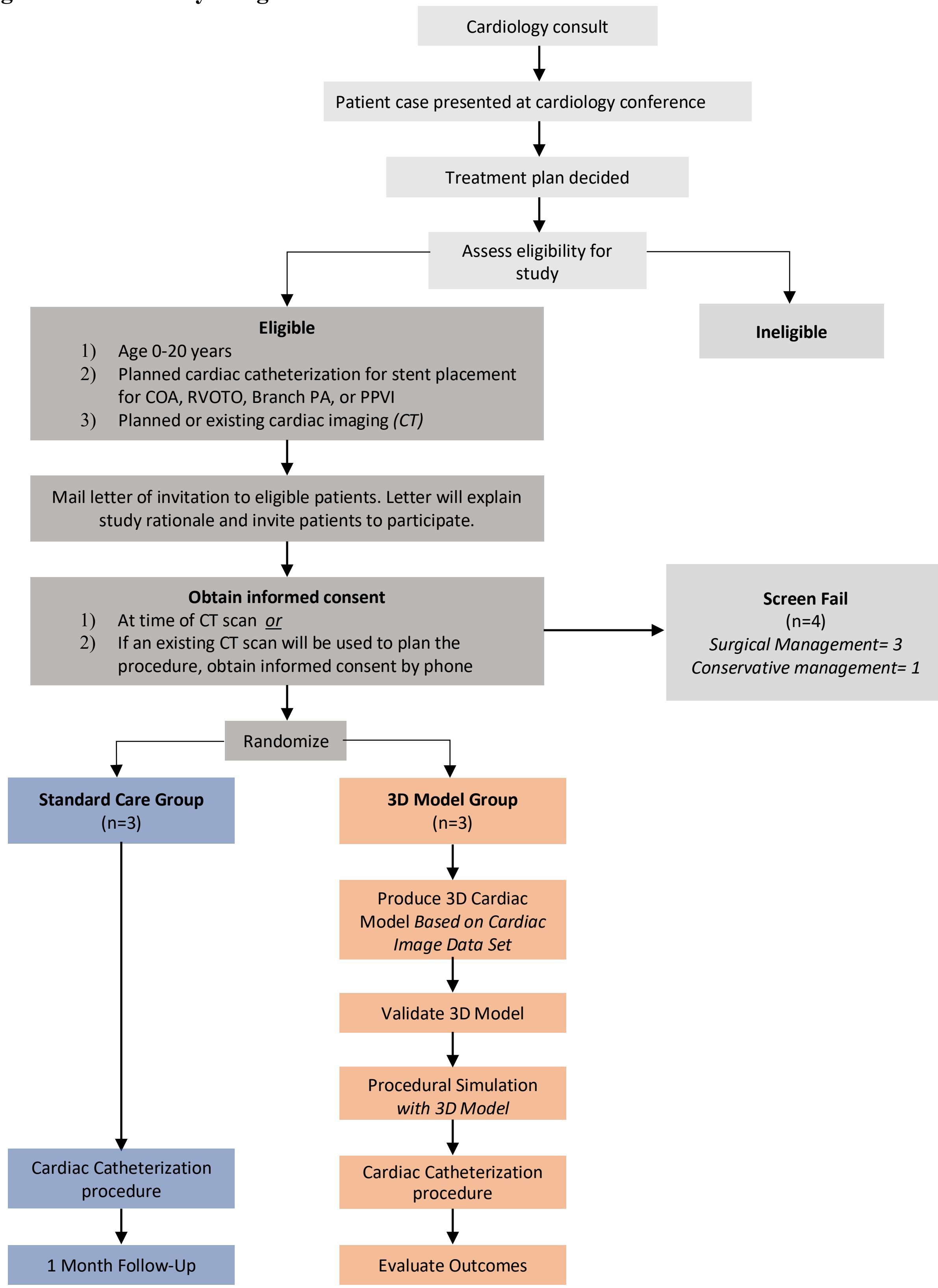


Figure 4.2 3D printed models in the cardiac catheterization laboratory
3D cardiac model used for IC planning and simulation (Panel A) under fluoroscopic projection (Panel B). Panel C shows a 3D cardiac model depicting the right ventricular outflow tract, during pre-procedural simulation of stent placement in the RVOT.

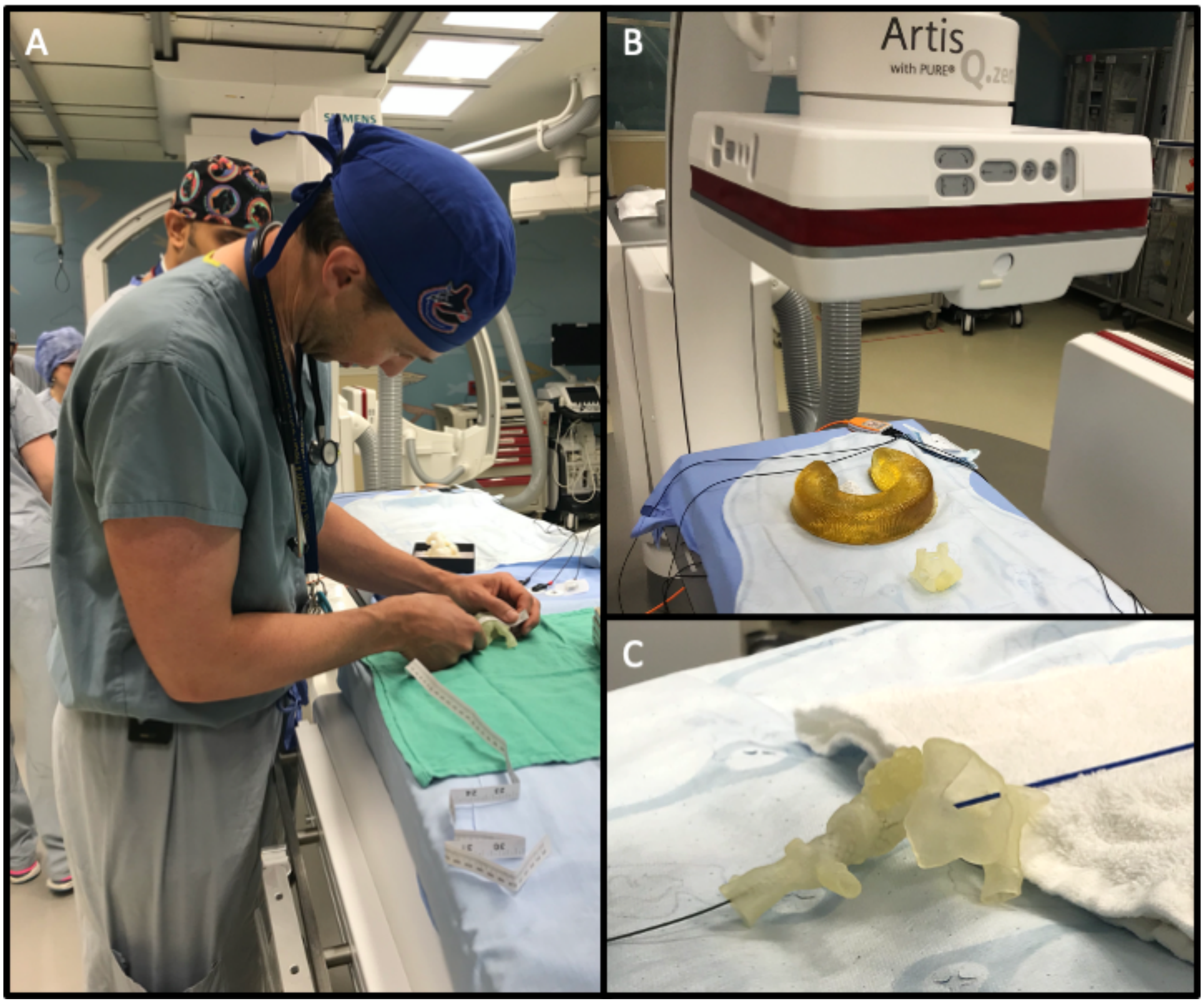


Figure 4.3 Stent placement for coarctation of the aorta
Illustration of pre-procedural IC stent placement simulation for coarctation of the aorta. Panel A: Pre-procedural CT of patient's aorta through the sagittal plane. Panel B: Pre-procedural x-ray angiography of patient's aorta. Panel C: X-ray angiography of patient's aorta following stent placement. Panel D: Pre-simulation photograph of 3D-CM. Panel E: Pre-simulation CT of 3D model. Panel F: Pre-simulation 3D rotational angiography of 3D model. Panel G: 3D rotational angiography of 3D model following stent placement. Panel H: Photograph of 3D model following stent placement.

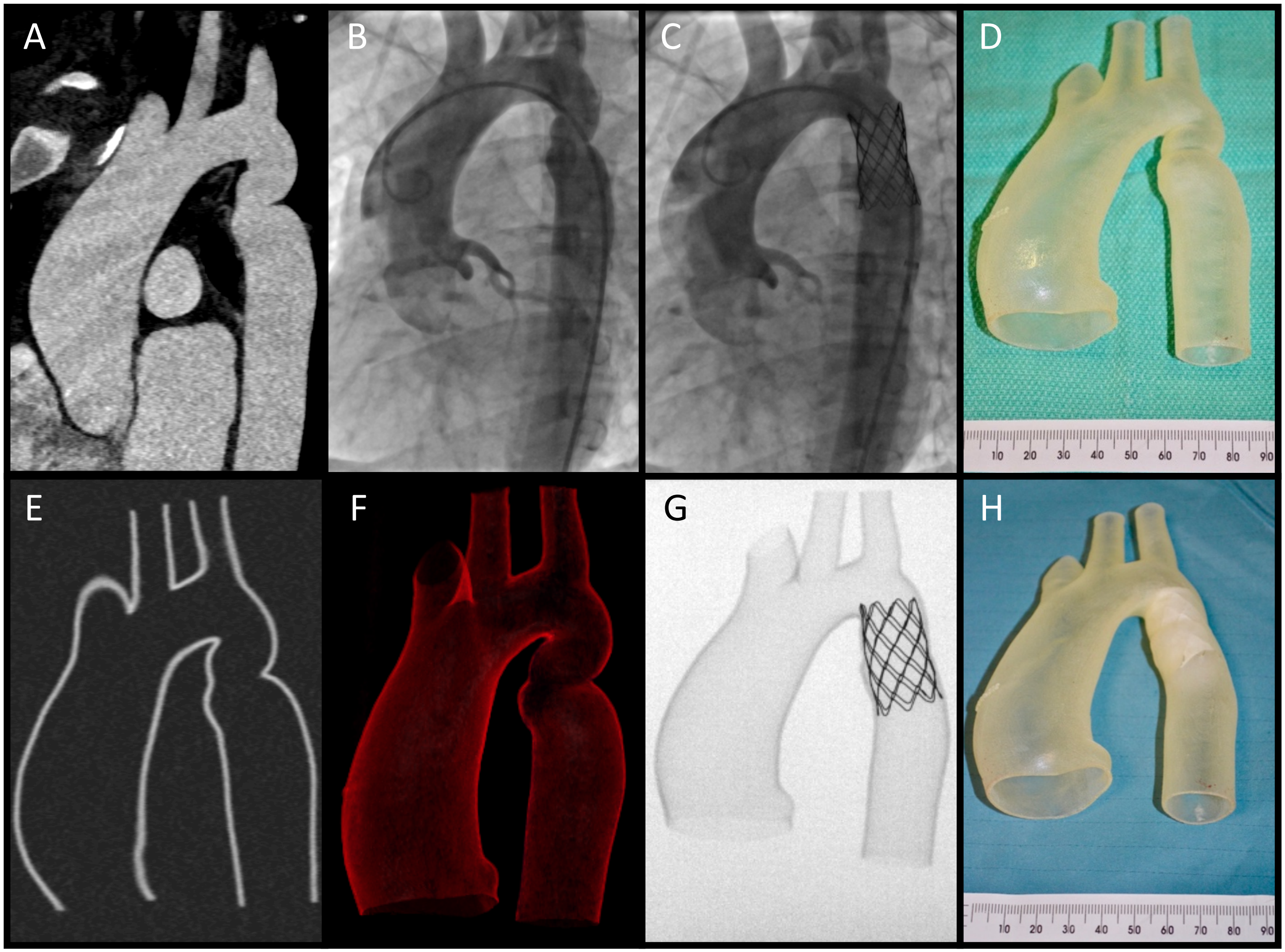


Figure 4.4 Agreement of 3D model and patient anatomy

Measurements from analogous sites on 3D models had excellent agreement with patient's anatomy on clinical imaging. Panel A: correlation of patient anatomy and 3D model measurements. Panel B: Bland-Altman plot of mean perimeter and perimeter difference of measured sites. All measures fell within the limits of agreement set at ± 1.96 SD.

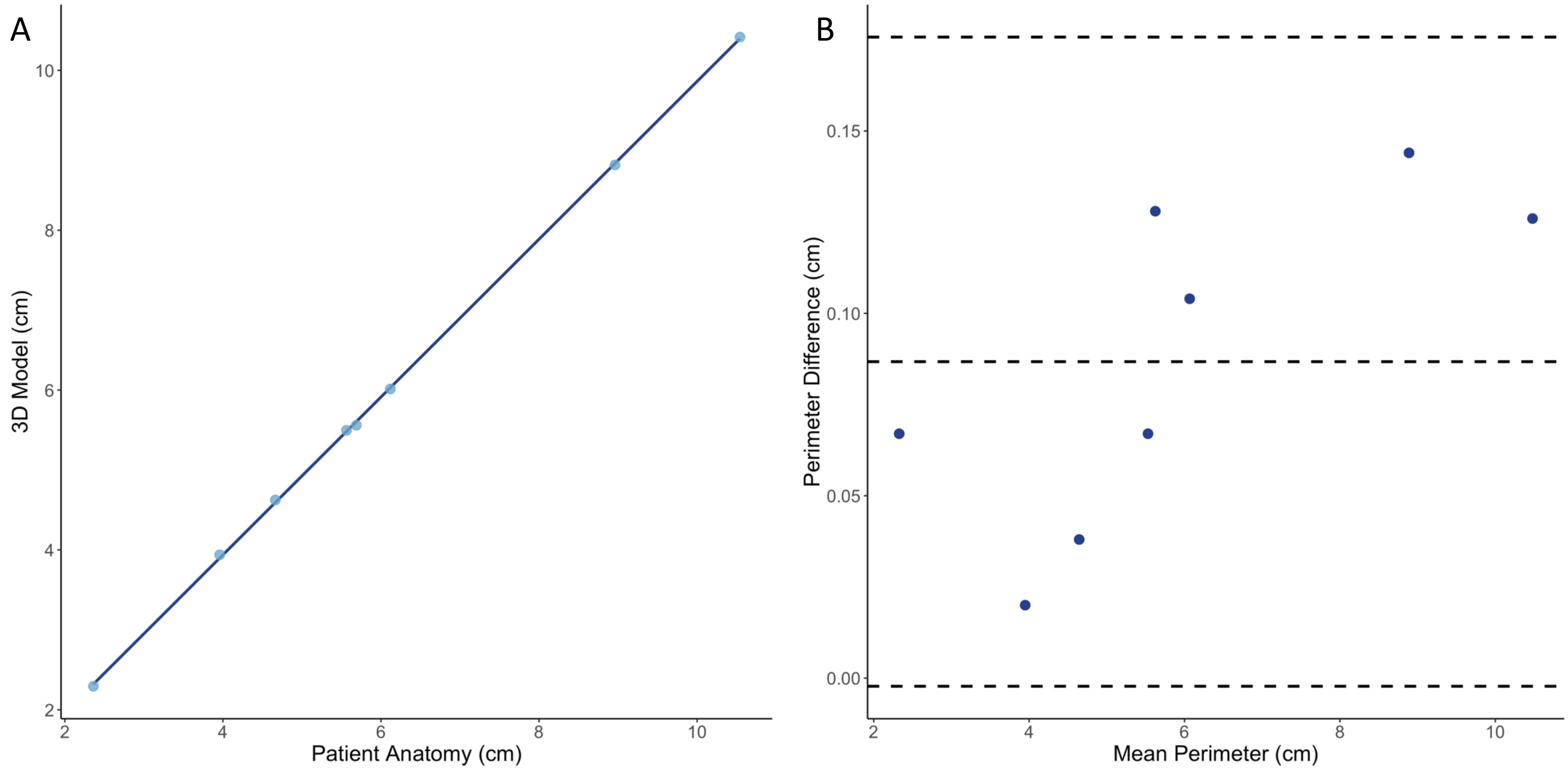


Table 4.1 Demographic and procedure related data

	Study Arm	
	Standard Care	3D Model
Demographic		
n	3	3
% female	100	100
Age (years), mean	14.6	10.9
Weight (kg), mean	67.3	43.0
Procedure Type		
CoA Stent Placement (n)	2	1
RVOT Stent Placement (n)	0	1
Branch Pulmonary Artery Stent Placement (n)	1	0
Percutaneous Pulmonary Valve Implantation (n)	0	1
Procedure Time (min), mean	90	87
Simulation Time (min), mean	-	16
Procedure Related Complications	none	none
Late Complications	none	none
Procedure Related Radiation Exposure		
DAP indexed to body weight ($\mu gy^2/kg$)	127.7	59.2
Total Radiation Dose (μgy^2)	669.3	192.4
Fluoroscopy Time (min)	28.3	18.6
Angiograms (n)	5.7	5.7

Table 4.2 Stent types

Stent size and type used during the interventional cardiac catheterization procedure and simulation, if applicable.

Patient ID	Diagnosis	Stent Size and Type		Change
		Simulation	Procedure	
1	CoA	CP Covered Stent *3.9 cm in length* *mounted on a 20 mm balloon*	CP Covered Stent *3.9cm in length* *mounted on a 20 mm balloon*	No
2	RVOT Obstruction	Genesis 3910 Transhepatic Biliary Stent *3.2 cm in length* *pre-mounted on a 10mm balloon*	Genesis XD2910 Stent *2.9 cm in length* *mounted on a 14mm balloon*	Yes
4	CoA	-	CP Covered Stent *3.4 cm in length* *mounted on a 18 mm balloon*	-
7	Pulmonary insufficiency	CP Bare Metal Stent *3.9 cm in length* *mounted on a 20 mm balloon*	CP Covered Stent *4.5 cm in length* *mounted on a 22mm balloon* Melody Transcatheter Pulmonary Valve	Yes
8	CoA	-	CP Covered Stent *3.2 cm in length* *mounted on a 20 mm balloon*	-
10	Branch PA Stenosis	-	CP Bare Metal Stent *2.2 cm in length* *mounted on a 12 mm balloon*	-

5 Evaluating the Use of 3D Cardiac Models in Undergraduate Medical Education: A Randomized, Controlled Trial

5.1 Introduction

Congenital heart disease (CHD) encompasses a heterogenous group of cardiac lesions that are present at birth and is the most common congenital malformation in newborns, with an incidence of about 1% (1). In CHD, cardiac structures can have complex three-dimensional (3D) spatial relationships, compared to normal anatomy. These 3D geometries may be difficult to conceptualize or interpret from images represented on a two-dimensional screen. Altered cardiac anatomy in CHD alters physiology (6).

3D printing is a novel technology that can be used to create a physical 3D model from a digital data set. Patient specific anatomical models can be manufactured with 3D printing by using a clinical imaging data set, such as computed tomography (CT), as the reference data file. To date, patient specific 3D printed cardiac models have been used in a variety of settings including procedural planning (12,38,40,70–73), patient and family education (29,30,72), and medical education (14,31,32,35,74,75). 3D printed cardiac models may be particularly useful for teaching CHD anatomy because users can physically handle and appreciate the lesions in 3D space.

Loke et al.'s study evaluating the utility of 3D printed cardiac models as a teaching aid for medical education have found that users reported higher satisfaction when learning with 3D models compared to 2D diagrams (35). Learners self-reported increased knowledge (14,37,74) and attain higher scores on objective knowledge assessments immediately after teaching sessions with a 3D cardiac model (32,34). It is unknown if 3D printed cardiac models used for medical education can improve knowledge retention over time. This study aimed to assess the impact of

3D printed cardiac models as teaching aids on learners' objective knowledge acquisition and retention of conotruncal abnormalities.

5.2 Materials and Methods

5.2.1 3D Modelling

Deidentified representative CT scans of unrepaired tetralogy of Fallot (TOF), d-Transposition of the Great Arteries (d-TGA), and normal cardiac anatomy were used to create two physical 3D models were created for each anatomy. Both TOF 3D models were hollow shell models to depict the ventricular septal defect and overriding aorta. For the normal and d-TGA anatomies one blood pool model and one hollow shell model were created. Hollow shell models were constructed from a flexible rubber-like material, Tango+ (Stratasys, Eden Prairie, MN). Blood pool models were constructed from a rigid polymer, Vero White (Stratasys, Eden Prairie, MN). For detailed 3D modelling methods please refer to *section 4.2.3.*

5.2.2 Congenital Heart Disease Workshop

Medical students in undergraduate year (UGY) 3 and 4 completing a clinical rotation at the Children's Heart Centre at BC Children's Hospital (BCCH) were invited to participate in a 1-hour standardized didactic workshop, in addition to their routine medical education. After providing informed consent to participate, medical students were group randomized, by workshop, in a 1:1 ratio, to attend the workshop with or without 3D printed cardiac models (3DM) as teaching aids (Fig. 5.1). The workshop was taught by a qualified pediatric cardiology clinician (SV) and focussed on the anatomy, physiology, and pathology of TOF and d-TGA, compared to normal cardiac anatomy.

For the 3DM group, the medical students participated in a 45-minute didactic lecture, followed by a 15-minute hands-on learning session with the 3D cardiac models. There were 2 normal cardiac anatomy models, 2 TOF models and 2 d-TGA models. Students were able to hold and physically manipulate the models. The workshop facilitator was available to highlight integral anatomical features and answer any questions. The control group attended the same 45-minute didactic lecture followed by a 15-minute question and answer period where the workshop facilitator was available to answer questions about CHD anatomy. This study was reviewed and approved by the BCCH Behavioural Research Ethics Board (H19-02607).

5.2.3 Knowledge Assessment

Participants' objective knowledge about normal cardiac, TOF and d-TGA anatomy and physiology was assessed prior to, immediately following and 10-days following the workshop. The knowledge assessment was a 23-point paper-based test consisting of multiple choice and fill-in-the-blank questions (Appendix D). The test was designed in consultation with an expert in pediatric cardiology (MCKH) experienced in writing medical examinations. The workshop instructor (SV) was blinded to the assessment questions in order to avoid bias in teaching. Participants were given 10 minutes to complete the assessment and instructed to leave answers blank if they did not know the answer to the question. The assessment also contained 3 questions asking participants to subjectively rate their knowledge about the normal heart, TOF and d-TGA on a 5-point Likert-type scale, where 1 was defined as "very low" and 5 was defined as "very high".

5.2.4 Survey

Participants were surveyed immediately following the workshop collecting demographic information on participants' age, educational background and previous cardiology or pediatric cardiology experience. Participants were asked to rate their agreement with the statement "This workshop helped me to learn about the normal heart TOF, or d-TGA" on a 5-point Likert-type scale. Additional space was provided for the participants to provide comments about the workshop. Participants in the 3DM group had an additional question to rate their agreement with the statement “The 3D printed cardiac models helped me to learn about the normal heart, TOF, or d-TGA” on a 5-point Likert-type scale. Descriptive statistics including counts, means, and percentages were calculated. Analysis was conducted using R (3.5.2, The R Foundation).

5.3 Results

Participants were recruited between January and March 2020. Due to the international COVID-19 pandemic a research curtailment order for all non-essential research was put in place at BCCH and all medical students were removed from clinical settings, including from the Children’s Heart Centre. Subsequently, all recruitment for this study was paused indefinitely, on March 16th, 2020. As a result, recruitment was lower than previously anticipated. Due to the small sample size, statistical analysis was not performed. Recruitment is planned to resume when the research curtailment at BCCH is lifted and normal clinical routine resumes at The Children’s Heart Centre.

7 participants were recruited, and 3 workshops were conducted with 3, 2, and 2 participants, respectively. Two workshops, and 5 participants were allocated to the 3D model group. All participants were male and enrolled in an MD program (Table 5.1).

Both the 3D model and control groups showed a similar trend of an increase in objective knowledge between the pre- and post- workshop assessment, followed by good knowledge retention over the 10-day follow-up period (Fig. 5.2). The control group had no change in mean knowledge assessment score over the 10-day retention period between the workshop and follow-up assessment. The 3D model group exhibited a less than 1% decrease in mean knowledge assessment score.

The control group exhibited greater baseline knowledge and higher knowledge assessment scores group at all time points, compared to the group taught with the 3D cardiac models. However, the 3D model group exhibited a greater increase in knowledge acquisition, with a mean 47% increase in assessment score, between the pre- and post-workshop knowledge assessments. The control group had a mean 37% increase in assessment score between the same time-points (Fig. 5.3).

All students agreed or strongly agreed that the workshop facilitated their learning about normal cardiac anatomy, TOF and d-TGA. Of the students allocated the to the 3D printing group, all agreed or strongly agreed that the 3D models facilitated their learning about TOF. 80% agreed or strongly agreed that the models facilitated learning about TGA and 60% agreed or strongly agreed the models facilitated learning about the normal anatomy (Table 5.2).

5.4 Discussion

CHD lesions may have complex 3D spatial relationships between intracardiac structures (76,77). These 3D geometries can be difficult for the learner or clinician to understand or conceptualize while represented on a flat screen (7). 3D models provide the learner or clinician the ability to view these 3D relationships in 3D space. In addition, the user can tactilely maneuver and examine the 3D model from various angles to fully appreciate the 3D anatomy, a task that is not possible from a 2D-image. The learners allocated to the 3D model group had the opportunity to hold and inspect each model. Anecdotally, many students, closely examined the relationships between the overriding aorta and both ventricles by inserting a probe, such as pencil or finger, through either side of the VSD. The students viewed the normal cardiac and d-TGA models to compare the connection of the ventricles and great arteries. The integration of visual and tactile learning provided by 3D models may be beneficial for users' understanding and comprehension. Our early results suggest that learners allocated to the 3D model group had greater increase in knowledge assessment score following the workshop, compared to those allocated to attend the same workshop without 3D cardiac models.

Over the past several decades there have been many developments in the treatment of CHD, including improvements in fetal detection, and innovations in surgical and interventional cardiology procedures (55,78). These factors have translated to an increased median life expectancy, well into adulthood, for patients living with CHD (55). In fact, there are now more adults, than children, living with CHD in Canada (2). Given the increase in adult CHD population, there is an increased likelihood that even physicians who are not cardiac-specialized will encounter patients with CHD during their practice. As such, CHD represents an important area of medical education for all medical students, regardless of their intended specialty.

Our study was conducted at a single centre, with a limited sample size. The small sample size prevented statistical analysis and hypothesis testing. As such, results should be interpreted with caution. Further recruitment to increase sample size is needed and is planned when the COVID-19-related research curtailment is lifted at BCCH.

5.5 Conclusions

This study demonstrated the feasibility of incorporating a CHD workshop with 3D cardiac models into the existing clinical rotation structure at The Children's Heart Centre at BCCH. The workshop was well received by learners, with a 100% recruitment rate. Our initial results suggest that 3D cardiac models may improve learners' knowledge acquisition of CHD such as TOF and d-TGA.

Figure 5.1 Study timeline.
Participants are randomized to the 3D model or control arm following informed consent. All participants complete a knowledge assessment at three time points (immediately prior to workshop, immediately following workshop, and 10-day post-workshop). Study participants take part in the same didactic workshop on CHD, with or without the use of 3D printed cardiac models.

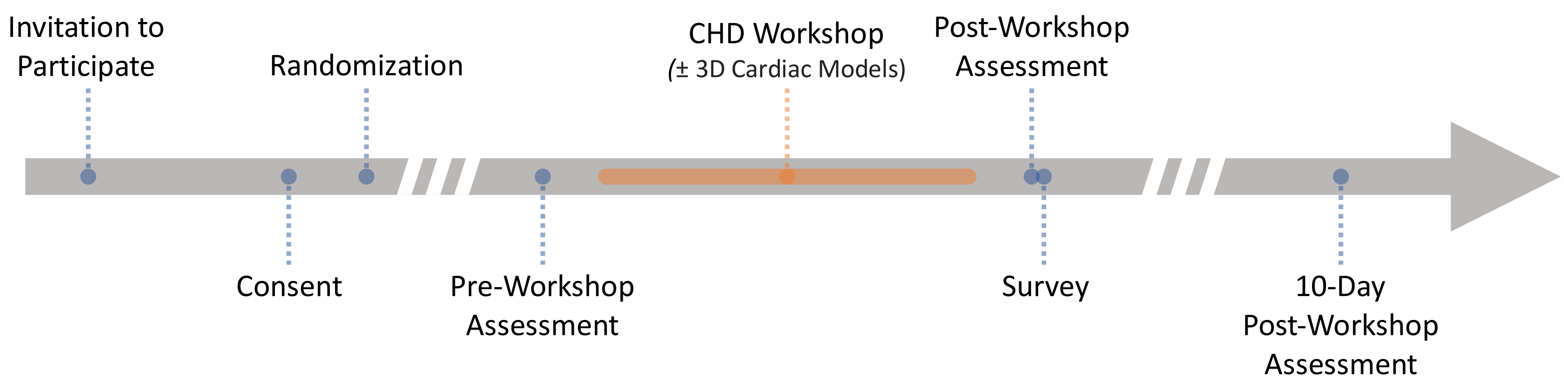


Figure 5.2 Objective knowledge over time
Participant objective knowledge as measured by score, in percentage, on the knowledge assessment.

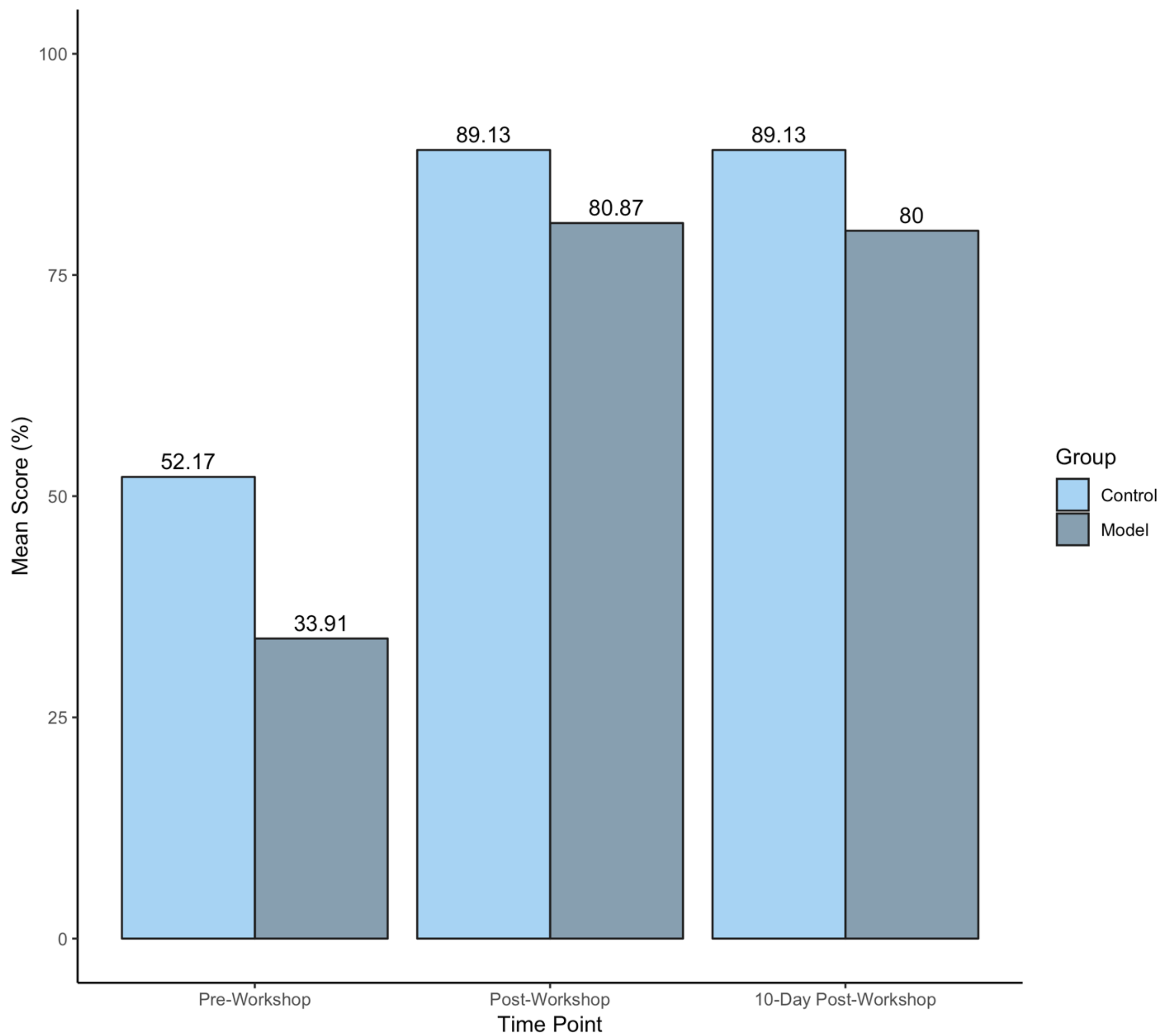


Figure 5.3 Knowledge acquisition

Knowledge acquisition between the pre- and post-workshop knowledge assessment. Change in score is reported in percentage.

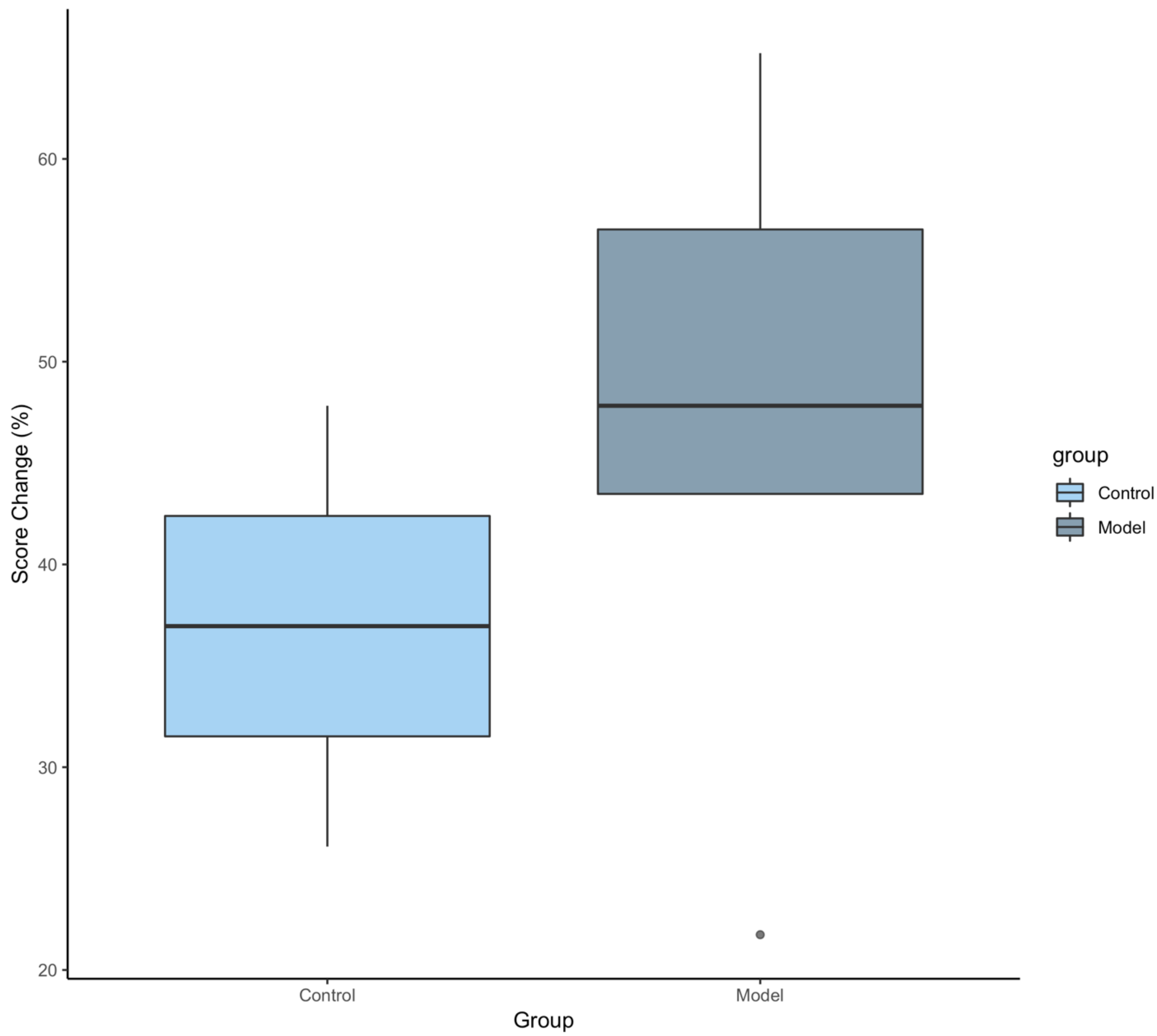


Table 5.1 Demographics
Participant's demographic data by study group.

	Control Group	3D Model Group
Students enrolled, n	2	5
Male, n (%)	2 (100)	5 (100)
Median age (years)	24.5	25
Highest Level of Education, n(%)		
Bachelor's	2 (100)	3 (60)
Master's	0 (0)	2 (60)
PhD	0 (0)	0 (0)
Program, n(%)		
MD	2 (100)	5 (100)
MD/PhD	0 (0)	0 (0)
Year of Training, n(%)		
UGY3	2 (100)	4 (80)
UGY4	0 (0)	1 (20)
Prior cardiology elective, n (%)	0 (0)	1 (20)

Table 5.2 Workshop and model ratings

Participants were asked to rate whether the workshop and 3D models facilitated their understanding about the normal heart, TOF and d-TGA on a 5-point Likert-type scale.

*Only participants allocated to the 3D Model group provided a rating on the effectiveness of the 3D models.

	Workshop			3D Models*		
	Normal Heart	TOF	d-TGA	Normal Heart	TOF	d-TGA
Rating 4-5 (most helpful), %	100	100	100	60	100	80
Rating 1-3 (least helpful), %	0	0	0	40	0	20
Mean Rating	4.4	5	5	4.0	4.4	4.3

6 Conclusions

Three-dimensional (3D) printing is a relatively new technology, with novel applications in congenital heart disease (CHD). Previous work (10,16) and our project (see Chapter 4) have demonstrated that 3D printing can produce highly accurate, anatomical cardiac models. The benefit of these models permitted appreciation of 3D relationships in 3D space, as opposed to on a two-dimensional screen. 3D cardiac models have been used in procedural planning, simulation, and education. However, our scoping review of the literature revealed there is a lack of scientifically robust reports in this area. Only 15% of studies report the use of control group. Most of the current body of literature pertains to anecdotal case reports and series that describe the use of 3D CHD models in practice. However, these reports do not define the magnitude or direction of impact that 3D cardiac models can have in the management of patients with CHD. There is a need for future large and controlled studies to determine the best applications for this technology. Our work has identified gaps in the literature, highlighted priority areas for future research (Table 2.2) and, sought to fill some of these gaps through randomized, controlled trials. Our pilot study assessing use of 3D cardiac models for interventional cardiology simulation is, to our knowledge, the first randomized controlled pilot study to use 3D cardiac models in pre-procedural simulation for interventional cardiology. The data suggest the benefit of pre-procedural simulation may be a reduction in procedure related radiation exposure. These initial findings have been used to inform a grant proposal for a large, multi-centre randomized controlled trial using the same procedures.

As a contemporary technology, 3D printing is still not accessible to all clinicians. In our international survey of pediatric cardiologists, we found that less than half of respondents had access to the technology. Access was dependent on location of the respondent and may be influenced by the healthcare funding sources where the clinician practices. We anticipate that the cost of this technology will decrease and access to the technology would increase over time permitting better access to 3D printing.

3D printing is a new and useful technology in CHD with applications and potential as both a clinical and educational modality. Our work has investigated the current applications of this technology and has laid a foundation for future robust scientific studies in the area of 3D printing technology for CHD.

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Appendix A

Example search strategy from MEDLINE.

▼ Search History (9) View Saved

	# ▲	Searches	Results	Type	Actions	Annotations
	1	exp Heart Defects, Congenital/	145734	Advanced	Display Results | More	
	2	("congenital heart malformation" or "congenital heart disease" or "congenital heart defect").mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]	27476	Advanced	Display Results | More	
	3	("patent ductus arteriosus" or "hypoplastic left heart syndrome" or "heart single ventricle" or "heterotaxy syndrome" or "dextrocardia" or "ebstein anomaly" or "heart left right shunt" or "heart septum defect").mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]	15808	Advanced	Display Results | More	
	4	1 or 2 or 3	157821	Advanced	Display Results | More	
	5	exp Printing, Three-Dimensional/	3576	Advanced	Display Results | More	
	6	("3D Printing" or "Three-Dimensional Printing" or "3D Printed Model" or "Three-Dimensional Printed Model" or "3D Model" or "Three-Dimensional Model" or "Rapid Prototyping" or "Additive Manufacturing").mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]	14594	Advanced	Display Results | More	
	7	("Stereolithography" or "Three Dimensional Printing" or "Three Dimensional Printed Model" or "Three Dimensional Model").mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]	5463	Advanced	Display Results | More	
	8	5 or 6 or 7	16386	Advanced	Display Results | More	
	9	4 and 8	195	Advanced	Display Results | More	

Contract

Appendix B

Summary table of studies included in final qualitative synthesis.

First Author	Title	Area(s) of Utilization	Lesion	Aim	Outcome Measures	Findings
Xu, J (2019)	Patient-Specific Three-Dimensional Printed Heart Models Benefit Preoperative Planning for Complex Congenital Heart Disease	1. Surgical Planning	1. PAPVC, ASD (± CoA) 2. TAPVC (supracardiac and infracardiac, intracardiac) ± ASD, PDA 3. TOF (with collateral vessels of descending aorta) 4. TOF, ASD 5. Right aortic arch, left carotid artery malformation PS, ASD, and PDA 6. TGA, IAA (type A), PDA, VSD, ASD, PS 7. DORV, VSD, PDA, CoA 8. Ascending aorta and aortic arch dysplasia, PDA, VSD, ASD 9. Severe PS, AS	1. To investigate the usefulness of 3D printed patient-specific heart models in the pre-surgical planning for complex congenital heart disease	1. Change in surgical approach and plan based on 3D printed models	1. Patient-specific 3D printed heart models are able to accurately provide the intracardiac anomalies prospectively before surgery 2. 3D printed heart models improve the operator's understanding of the complex spatial structure. Since different anatomical variants require different surgical approaches and treatment strategies, the 3D models could help cardiologists make appropriate decisions before surgery
Sahayaraj, R (2019)	3D Printing to Model Surgical Repair of Complex Congenitally Corrected Transposition of the Great Arteries	1. Surgical Planning	1. ccTGA, PA	1. To describe the utility of a patient-specific 3D heart model in surgical planning for complex CHD.	No pre-defined outcome measures	1. Intra-operative findings were consistent with the 3D model 2. The model helped to determine the feasibility of biventricular repair, delineate access route to close the VSD, and plan for optimal position of the conduit.
Carberry, T (2019)	Fontan Revision: Pre-surgical Planning Using Four Dimensional (4D) Flow and Three-Dimensional (3D) Printing	1. Surgical Planning	1. Heterotaxy syndrome, DORV, hypoplastic right ventricle, PS, interrupted inferior vena cava with left azygous continuation, left superior vena cava, and right aortic arch	1. To assess the usefulness of a patient-specific 3D printed cardiac model of complex congenital heart disease in pre-surgical planning of a Fontan revision	1. Pulmonary vein blood flow pre and post-op measured by 4D flow MRI	1. The 3D model helped surgeons understand the anatomic relationship between hepatic vein, left azygos vein, esophagus and surrounding structures. 2. The delineation of anatomy guided surgical planning.

Ryan, J (2018)	3D Printing for Congenital Heart Disease: A Single Site's Initial Three-Year Experience	1. Surgical Planning	1. PA, VSD 2. TOF (PA and absent pulmonary valve) 3. DORV (TGA variant) 4. TA 5. Vascular rings (not specified) 6. Single Ventricle (not specified)	1. To assess the impact of 3D printing on a single healthcare institution 2. To understand what metrics serve as ideal endpoints for future studies 3. To inform the creation of a clinical trial with 3D printing as a experimental arm	1. Operative time (in minutes) 2. 30 day readmission (Y/N) 3. 30 day mortality (Y/N)	The use of 3D printed cardiac models led to: 1. Reduced operative time (did not reach significance) 2. No difference in 30 day mortality 3. No difference in 30 day readmission. 4. Cardiac surgeons found that the models enhanced their efficiency to execute surgical repair.
Mendez, A (2018)	Apical Muscular Ventricular Septal Defects: Surgical Strategy Using Three-Dimensional Printed Model	1. Surgical Planning	1. Multiple apical VSD's in the trabecular septum, perimembranous VSD, and ASD	1. To determine a pre-operative plan for VSD repair using 3D printed model	No pre-defined outcome measures	1. The 3D model guided the selection of appropriate patch size and optimal location for patch placement.
Zhao, L (2018)	Three-Dimensional Printing Enhances Preparation for Repair of Double Outlet Right Ventricular Surgery	1. Surgical Planning	1. DORV	1. To assess the usefulness of 3D printed cardiac models for surgical planning of DORV	1. Operative Time 2. Cardiopulmonary Bypass Time 3. Aortic Cross Clamping Time 4. Mechanical Ventilation Time 5. Intensive Care Unit Stay	1. Patients in the 3D printing group had shorter aortic cross-clamp time and cardiopulmonary bypass time than patients in the control group but this was not statistically significant. 2. Patients with 3D printed models had significantly lower mechanical ventilation time and significantly shorter intensive care unit time than patients in the control group.
Juaneda, I (2018)	Three-Dimensional Printing Model in Double-Outlet Right Ventricle to Simplify Intraventricular Repair	1. Surgical Planning	1. DORV Fallot-type, moderate valvar and subvalvular PS	1. To assess the feasibility of using patient-specific 3D model of DORV for surgical simulation and planning	No pre-defined outcome measures	1. The 3D printed model accurately demonstrated the cardiac anatomy including the relationship between the VSD and aorta and successfully guided the surgical approach.
Moore, R (2018)	Three-Dimensional Printing in Surgical Planning: A Case of Aortopulmonary Window with Interrupted Aortic Arch	1. Surgical Planning	1. APW, IAA (type A), and PDA	1. To assess the usefulness of using a patient-specific 3D printed model of APW associated with IAA to guide surgical planning and simulation	No pre-defined outcome measures	1. The 3D printed model delineated the extent of proximity of APW to the interrupted arch and size of APW and successfully guided surgical planning.

Valverde, I (2017)	Three-Dimensional Printed Models for Surgical Planning of Complex Congenital Heart Defects: An International Multicentre Study	1. Surgical Planning	1. DORV 2. TGA 3. Univentricular heart physiology (not specified) 4. VSD 5. Criss-cross heart 6. LVOT obstruction 7. Heterotaxy syndrome 8. Discordant AV and VA connections	1. To evaluate the usefulness of patient-specific 3D printed cardiac models to guide surgical planning of complex CHD.	1. Change in procedural plan 2. Satisfaction with utilizing 3D printed cardiac models 3. Perception of usefulness of 3D models	1. In 19 of the 40 complex cases, consideration of a 3D model redefined surgical management. 2. Cardiologists and cardiac surgeons perceive 3D models as useful for medical education and facilitating communication with colleagues and patients.
Smith, M (2017)	The Role of 3D Printing in Preoperative Planning for Heart Transplantation in Complex Congenital Heart Disease	1. Surgical Planning	1. Situs inversus dextrocardia, TGA, single atrium, DORV, TAPVC to the left SVC	1. To describe the use of a 3D printed model to guide surgical planning of heart transplantation in a patient with congenital heart disease.	No pre-defined outcome measures	1. The model helped to determine required length of donor ascending aorta, SVC and brachiocephalic vein during harvest. 2. The model allowed recognition of anomalous drainage of pulmonary veins and avoided intraoperative dissection and exploration.
Vodiskar, J (2017)	Using 3D Physical Modeling to Plan Surgical Corrections of Complex Congenital Heart Defects	1. Surgical Planning	1. DORV, TGA, VSD, CoA, aortic arch hypoplasia 2. IAA, VSD, hypoplastic aortic valve, small ascending aorta 3. DORV, side to side malposition of the great arteries, subpulmonary VSD	1. To assess the usefulness of patient-specific 3D printed heart models for surgical planning in complex congenital heart disease.	No pre-defined outcome measures	1. The models helped to decide on surgical approach. 2. The models assisted pre-surgical decision of vascular prosthesis size. 3. The models' limitation was that fine intracardiac structures such as valves and chordae could not be delineated.
Kappanayil, M (2017)	Three-Dimensional-Printed Cardiac Prototypes Aid Surgical Decision-Making and Preoperative Planning in Selected Cases of Complex Congenital Heart Diseases: Early Experience and Proof of Concept in a Resource-Limited Environment	1. Surgical Planning	1. DORV, VSD, side by side great arteries 2. ccTGA, PA, large inlet VSD 3. Crisscross atrioventricular connection, VSD, DORV, D-TGA, CoA, PDA 4. Criss cross atrioventricular connections, VSD, L-TGA 5. Multiple VSD's, PA	1. To describe the use of 3D printed cardiac models for surgical decision making and surgical planning in cases of CHD.	No pre-defined outcome measures	1. The 3D printed cardiac models can be utilized to plan surgical procedures including surgical approach, site of cardiotomy, selecting size of patch, and sites of conduit placement

Fernandez Carbonell, A (2017)	Correcting Congenital Heart Diseases with 3D Models	1. Surgical Planning	1. HLHS	1. To describe a case where a 3D printed heart model of HLHS was used to plan cardiac surgery.	No pre-defined outcome measures	1. Using a 3D printed cardiac model, surgeons were able to plan corrective surgery (atrio-pulmonary connection with intracardiac fenestrated tunnel) and avoid transplantation.
Hadeed, K (2016)	Three-Dimensional Printing of a Complex CHD to Plan Surgical Repair	1. Surgical Planning	1. DOLV (pulmonary artery arose from left ventricle, PS, overriding aorta, subaortic VSD)	1. To describe a case where a 3D printed heart model of DOLV was used to plan surgical repair	No pre-defined outcome measures	1. The 3D printed cardiac model of a patient with DOLV offered better understanding of the spatial relationships between great arteries and intra-cardiac structures and assisted with surgical planning.
Farooqi, K (2016)	Use of a Three Dimensional Printed Cardiac Model to Assess Suitability for Biventricular Repair	1. Surgical Planning	1. DORV with non-committed VSD status post bidirectional Glenn anastomosis	1. To describe a case where a 3D printed cardiac model of DORV was used in pre-surgical planning and decision making.	No pre-defined outcome measures	1. The spatial information gained from the 3D model led the surgical team to decide on a biventricular repair (over Fontan palliation).
Farooqi, K (2015)	Use of 3-Dimensional Printing to Demonstrate Complex Intracardiac Relationships in Double-Outlet Right Ventricle for Surgical Planning	1. Surgical Planning	1. Dextrocardia, DORV and supratricuspid ring status post pulmonary artery banding	1. To describe the utility of a 3D printed model in surgical planning for a patient with DORV.	No pre-defined outcome measures	1. The 3D model demonstrated intracardiac anatomy and the spatial relationship between the great vessels, ventricles and VSD. With the model, a double-switch procedure was decided and performed.
Ma, X (2015)	Clinical Application of Three-Dimensional Reconstruction and Rapid Prototyping Technology of Multislice Spiral Computed Tomography Angiography for the Repair of Ventricular Septal Defect of Tetralogy of Fallot	1. Surgical Planning	1. TOF	1. To compare intra-operative measurements of cardiac structures with analogous sites on 3D printed models.	1. Size of VSD (mm)	1. The intra-operative use of 3D models assisted with surgical planning by allowing to select the appropriate patch size for VSD closure.
Valverde, I (2015)	Three-Dimensional Patient-Specific Cardiac Model for Surgical Planning in Nikaidoh Procedure	1. Surgical Planning	1. TGA, subaortic VSD, PS status post Blalock-Taussig shunt	1. To describe the use of patient-specific 3D printed model of TGA, VSD and PS to guide surgical planning.	No pre-defined outcome measures	1. The 3D model corroborated the decision for Nikaidoh procedure and facilitated technical planning.

Schmauss, D (2015)	Three-Dimensional Printing in Cardiac Surgery and Interventional Cardiology: A Single-Centre Experience	1. Surgical Planning	1. Right aortic arch with abberant left subclavian artery 2. Subpulmonary VSD 3. HLHS 4. PA and hypoplastic right ventricle	1. To describe how patient specific 3D cardiac models were used at a single institution.	No pre-defined outcome measures	1. The intra-operative use of patient-specific 3D cardiac models assisted with surgical planning and transplant planning for cases of previously corrected CHD.
Sodian, R (2008)	Pediatric Cardiac Transplantation: Three-Dimensional Printing of Anatomic Models for Surgical Planning of Heart Transplantation in Patients with Univentricular Heart	1. Surgical Planning	1. HLHS 2. PA and hypoplastic right ventricle	1. To assess the feasibility of using patient-specific 3D printed models of patients with univentricular physiology for surgical planning of cardiac transplantation	No pre-defined outcome measures	1. The intra-operative use of patient-specific 3D printed cardiac models assisted with optimizing the surgeon's orientation and with planning cardiac transplantation.
Mottl-Link, S (2008)	Physical Models Aiding in Complex Congenital Heart Surgery	1. Surgical Planning	1. Dextrocardia, TGA, PA, large VSD, ASD	1. To describe the utility of a 3D printed cardiac model for surgical planning and intra-operative orientation.	No pre-defined outcome measures	1. The blood pool model assisted with pre-surgical planning. 2. The myocardial model improved intra-operative orientation during surgery.
Sodian, R (2007)	Stereolithographic Models for Surgical Planning in Congenital Heart Surgery	1. Surgical Planning	1. Right aortic arch with aberrant left subclavian artery 2. Subpulmonary VSD	1. To describe the use of patient-specific 3D printed models of vascular ring and VSD for choosing treatment strategies, surgical planning of corrections, and intra-operative orientation	No pre-defined outcome measures	1. The 3D printed model assisted with surgical planning and deciding on surgical approach. 2. The intra-operative use of the model improved the surgeon's orientation.

Riesenkampff, E (2009)	The practical clinical value of three-dimensional models of complex congenitally malformed hearts	1. Surgical Planning	1. DORV ± LVOTO, RVOTO, CoA, multiple VSDs, valvar PS, cAVSD 2. Swiss-cheese VSDs, RVOTO, valvar PS 3. TOF, severe PS, VSD 4. ccTGA, pulmonary atresia, VSD 5. TGA, valvar PS, VSD 6. AVSD, unbalanced ventricles, subvalvar PS 7. ccTGA, pulmonary atresia, VSD 8. Multiple muscular VSDs, TAPVC, discordant atrioventricular connections, infundibular and valvar PS	1. To assess the usefulness of patient specific 3D cardiac models for operative planning	No pre-defined outcome measures	1. The models facilitate and improve understanding of pathology and provide decisive information about spatial position, size and relationship of structural anomalies.
Jivanji, S (2019)	Novel Use of a 3D Printed Heart Model to Guide Simultaneous Percutaneous Repair of Severe Pulmonary Regurgitation and Right Ventricular Outflow Tract Aneurysm	2. Interventional Cardiology Planning 3. Interventional Cardiology Simulation	9. PA, VSD, multiple aortopulmonary collaterals	2. To assess the feasibility of trans-catheter closure of RVOT aneurysm through simulation of the procedure on a 3D printed cardiac model.	1. The feasibility of transcatheter closure of RVOT aneurysm, as measured by the success of the procedure	2. Using 3D printed cardiac models for simulation helped to determine positioning of the valve and aneurysm device 3. Simulation led cardiologists to proceed with procedure 4. Simulation confirmed the feasibility of the procedure
Shijo, T (2018)	Stent Grafting Simulation Using a Three-Dimensional Printed Model for Extensive Aortic Arch Repair Combined with Coarctation	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. CoA with severe AS and multiple thoracic aortic aneurysms	1. To describe simulated thoracic endovascular aortic repair using a patient specific 3D model.	No pre-defined outcome measures	1. The 3D printed models assisted with simulation, planning and feasibility of thoracic endovascular aortic repair by predicting necessary stent size.

Imai, M (2018)	Successful Catheter Treatment Using Pre-Operative 3D Organ Model Simulation for Atrial Septal Defect With Dextrocardia and Interrupted Inferior Vena Cava to the Superior Vena Cava	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Situs inversus, Dextrocardia, and interrupted inferior vena cava (IVC) with azygous continuation and ASD	1. To describe the use of a patient-specific 3D printed model of ASD in guiding catheter assisted device closure	No pre-defined outcome measures	Pre-procedural simulation predicted successful ASD closure
Thakkar, A (2018)	Transcatheter Closure of a Sinus Venosus Atrial Septal Defect Using 3D Printing and Image Fusion Guidance	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Sinus venosus ASD and PAPVC	1. To assess the feasibility of using a patient-specific 3D printed model of sinus venous ASD in simulating and planning catheter-assisted device closure	No pre-defined outcome measures	1. A patient specific 3D printed model of sinus venous ASD assisted with simulation and planning of catheter assisted device closure in a complex patient not suitable for conventional surgical therapy.
So, K (2017)	Using Multimaterial 3-Dimensional Printing for Personalized Planning of Complex Structural Heart Disease Intervention	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. ASD	1. To describe the utility of a 3D printed cardiac model in ASD closure planning and simulation and the selection of optimal occluder size.	No pre-defined outcome measures	1. The patient-specific 3D cardiac model assisted with pre-procedural planning and selection of the appropriate occluder size for ASD closure.
Luo, H (2017)	Three-Dimensional Printing Model-Guided Percutaneous Closure of Atrial Septal Defect	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. ASD	1. To describe a case where a 3D printed cardiac model was used to plan ASD closure.	No pre-defined outcome measures	1. A patient-specific cardiac model allowed the selection of the appropriate-sized occluder for ASD closure. ASD.
Qiu, X (2017)	Feasibility of Device Closure for Multiple Atrial Septal Defects Using 3D Printing and Ultrasound-Guided Intervention Technique	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Multiple ASD's	1. To investigate the feasibility of trans-catheter closure of multiple ASD's monitored by trans-thoracic echocardiography under the guidance of a 3D printed heart model.	1. Selection of the most appropriate occlusion program through the simulation test on the model.	1. The patient-specific cardiac model of ASD provided a useful reference for trans-catheter device closure through ultrasound guided intervention technique. 2. This technique was successful in all 21 patients using a single atrial septal occluder. Mild residual shunt was found in 5 patients in the immediate postoperative period. Three of them disappeared during postoperative follow-up.

Pluchinotta, F (2017)	Stenting Complex Aortic Coarctation: Simulation in a 3D Printed Model	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. CoA	1. To describe the use of a 3D printed cardiac model for simulation of stent placement with deployment of 2 balloons.	No pre-defined outcome measures	1. A patient-specific 3D cardiac model of CoA assisted with planning and simulation of the cardiac catheterization intervention, stent length selection and stent position.
Ratnayaka, K (2017)	First-in-Human Closed-Chest Transcatheter Superior Cavopulmonary Anastomosis	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Dextrocardia, "criss-cross" AV connections, hypoplastic tricuspid valve and right ventricle, VSD, and subvalvar and valvar PS	1. To demonstrate the feasibility of a catheter-only, closed-chest, large-vessel anastomosis guided by simulation on a patient-specific 3D printed heart model.	No pre-defined outcome measures	1. A patient-specific 3D printed cardiac model assisted with planning and simulation of the first in-human trans-catheter large vessel anastomosis.
Ghisiawan, N (2016)	The Use of a Three-Dimensional Print Model of an Aortic Arch to Plan a Complex Percutaneous Intervention in a Patient with Coarctation of the Aorta	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. CoA status post stent and pseudoaneurysm	1. To describe the use of a patient-specific 3D printed cardiac model to plan and simulate percutaneous interventions.	No pre-defined outcome measures	1. A patient-specific 3D cardiac model assisted with simulation and planning of the percutaneous procedure and determining optimal stent position.
Wang, Z (2016)	Three-Dimensional Printing-Guided Percutaneous Transcatheter Closure of Secundum Atrial Septal Defect with Rim Deficiency: First-in-Human Series	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Secundum ASD	1. To determine if a 3D printed heart model would aid in percutaneous trans-catheter closure of secundum ASD by pre-procedural simulation.	No pre-defined outcome measures	1. Patient specific 3D Printed models of secundum ASD assisted with simulation and planning of percutaneous device closure
Chaowu, Y (2016)	Three-Dimensional Printing as an Aid in Transcatheter Closure of Secundum Atrial Septal Defect With Rim Deficiency: In Vitro Trial Occlusion Based on a Personalized Heart Model	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Secundum ASD with rim deficiency	1. To describe the use of a 3D printed heart model for simulation and the selection of the appropriate occluder size in preparation for trans-catheter device closure.	No pre-defined outcome measures	1. A patient-specific 3D cardiac model assisted with simulation of trans-catheter and allowed the selection of appropriate occluder size.

Phillips, A (2016)	Development of a Novel Hybrid Strategy for Transcatheter Pulmonary Valve Placement in Patients Following Transannular Patch Repair of Tetralogy of Fallot	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. TOF status post transannular patch repair	1. To utilize 3D printed cardiac models to develop a hybrid implant procedure for patients with irregular shaped RVOT requiring pulmonary valve replacement.	No pre-defined outcome measures	1. Patient-specific 3D cardiac models helped to successfully design a hybrid approach to remodel the RVOT in TOF patients.
Valverde, I (2015)	3D Printed Models for Planning Endovascular Stenting in Transverse Aortic Arch Hypoplasia	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. Hypoplastic transverse aortic arch, previous CoA status post end to end anastomosis	1. To describe the utility of a 3D printed model in simulation and planning of endovascular stenting in transverse aortic arch hypoplasia.	1. Size of model (mm) at specific locations	1. A patient-specific 3D cardiac model assisted with simulation of stent placement and procedural planning. 2. Simulation was used to predict best fluoroscopic projection angles used during procedure. 3. The models more accurately depicted patient's anatomy, compared to X ray angiography and MRI.
Olivieri, L (2014)	3D Heart Model Guides Complex Stent Angioplasty of Pulmonary Venous Baffle Obstruction in a Mustard Repair of D-TGA	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. D-TGA status post mustard procedure	1. To describe the utility of a 3D printed cardiac model in simulation and planning of a stent angioplasty in pulmonary venous baffle obstruction.	No pre-defined outcome measures	1. A patient-specific cardiac model assisted with simulation of catheter access and stent deployment prior to the procedure.

Matsubara, D (2019)	A Patient-Specific Hollow Three-Dimensional Model for Simulating Percutaneous Occlusion of Patent Ductus Arteriosus	1. Interventional Cardiology Planning 2. Interventional Cardiology Simulation	1. PDA	1. To determine if simulation of PDA closure via cardiac catheterization with patient specific 3D cardiac model prior to procedure can reduce procedure and fluoroscopy time.	1. Change in procedural route or device change 2. Change in procedural/fluoroscopic time and amount of contrast medium used 3. Procedural success vs failure or complications as measured by adequate fit of the device and disappearance of ducal flow after device insertion 4. Physicians' understanding of the procedures as measured by questionnaire	1. Patient-specific 3D printed models enabled pre-procedural selection of the PDA closure device and its size. 2. In the study group using 3D models, compared with the control group, the fluoroscopic and total procedural times were shorter. 3. 3D models improved physician understanding of the procedures.
Smerling, J (2019)	Utility of 3D Printed Cardiac Models for Medical Student Education in Congenital Heart Disease: Across a Spectrum of Disease Severity	1. Medical Education	1. PS 2. ASD 3. CoA 4. D-TGA 5. HLHS	1. To assess the use of 3D cardiac models in medical education of CHD lesions.	1. Self-assessed perceived knowledge (pre and post-workshop) assessed by a survey	1. Students' understanding of CHD lesions increased significantly with the use of 3D models and there was a positive association between the degree of complexity of CHD and the extent of increase in knowledge
White, S (2018)	Utility of Three-Dimensional Models in Resident Education on Simple and Complex Intracardiac Congenital Heart Defects	1. Medical Education	1. VSD (perimembranous, muscular, outlet) 2. TOF	1. To explore the utility of 3D models of simple and complex intracardiac lesions in resident education in a didactic lecture.	1. Pre and post-workshop score on an objective and a subjective knowledge test.	1. The incorporation of 3D printed models of CHD improves residents' understanding of TOF but not simple VSD's.

Su, W (2018)	Three-Dimensional Printing Models in Congenital Heart Disease Education for Medical Students: A Controlled Comparative Study	1. Medical Education	1. VSD (subarterial, membranous, and muscular types)	1. To determine if teaching medical students during a didactic session with 3D printed cardiac models yields higher results on an objective test than a didactic session without 3D printed cardiac models.	1. Academic performance as measured by objective multiple choice tests on VSD (using likert-type questionnaire and multiple choice	1. 3D printed cardiac models provide better structural conceptualization when used for teaching medical students
Loke, Y (2017)	Usage of 3D Models of Tetralogy of Fallot for Medical Education: Impact on Learning Congenital Heart Disease	1. Medical Education	1. TOF	1. To assess if teaching TOF with a 3D printed model will improve the learner's knowledge acquisition and overall satisfaction compared to learning with 2D drawings during a teaching session.	1. Knowledge acquisition as measured by comparing pre and post-session knowledge test scores. 2. Learner satisfaction and self-efficacy ratings as measured by questionnaires filled out by the residents after the teaching sessions.	1. Learners taught with 3D models reported higher learner satisfaction compared to learners taught with 2D diagrams. 2. There was no difference in knowledge acquisition between the 3D model or 2D diagram groups.
Biglino, G (2017)	Use of 3D Models of Congenital Heart Disease as an Education Tool for Cardiac Nurses	1. Medical Education	1. TGA status post repair 2. CoA 3. TOF 4. PA with intact ventricular septum 5. HLHS status post Norwood 6. HLHS status post Glenn 7. HLHS status post Fontan	1. To assess the feasibility of using 3D models of CHD for training nurses 2. To evaluate the nurses' perspectives on 3D models 3. To identify improvements to optimize the use of 3D models for training	1. Perceived usefulness of the course in improving learning of CHD 2. The most useful attributes of the model as measured by a questionnaire	1. Patient-specific 3D printed cardiac models can be helpful to train nurses. Models helped with learning experience and had more information than diagrams.

Olivieri, L (2016)	"Just-In-Time" Simulation Training Using 3-D Printed Cardiac Models After Congenital Cardiac Surgery	1. Medical Education	1. HLHS, TAPVC 2. Supravalvar AS 3. DORV, hypoplastic stenotic aortic valve, hypoplastic arch 4. AS and AI 5. PAPVC 6. Left pulmonary artery sling 7. RVOT obstruction 8. Truncal Valve regurgitation 9. Vascular ring (not specified) 10. TGA, VSD, PS	1. To evaluate the effects of clinician expertise and case complexity on the application of postoperative care simulation training in the cardiac intensive care setting.	1. Participant perception of the impact of the training session on the understanding of complex CHD as measured by a Likert-type scale questionnaire	1. The 3D heart models can be used to enhance congenital cardiac critical care via simulation training of multidisciplinary intensive care teams. Benefit may be dependent on provider type and case complexity.
Costello, J (2015)	Incorporating Three-Dimensional Printing into a Simulation-Based Congenital Heart Disease and Critical Care Training Curriculum for Resident Physicians	1. Medical Education	1. VSD (Infundibular, membranous, inlet, muscular and atrioventricular types)	1. To evaluate if 3D printed heart models can be effectively incorporated into a simulation-based CHD and critical care training curriculum for Pediatric resident physicians.	1. Knowledge acquisition 2. Knowledge reporting 3. Structural conceptualization As measured by Likert-type questionnaire pre and post seminar	1. Pediatric residents had improvement in the areas of knowledge acquisition, knowledge reporting, and structural conceptualization of VSD's, as well as improvement in the ability to describe and manage postoperative complications after completing a simulation training session with 3D printed cardiac models.
Costello, J (2014)	Utilizing Three-Dimensional Printing Technology to Assess the Feasibility of High-Fidelity Synthetic Ventricular Septal Defect Models for Simulation in Medical Education	1. Medical Education	1. VSD (infundibular, membranous, inlet, muscular, and atrioventricular types)	1. To assess the feasibility of utilizing 3D printed models of VSD to create a novel, simulation-based educational curriculum for premedical and medical students.	1. Knowledge acquisition 2. Knowledge reporting 3. Structural conceptualization As measured by Likert-type questionnaire pre and post seminar	1. Students had significant improvement in knowledge acquisition, knowledge reporting and structural conceptualization after attending a workshop with 3D models.
Balegadde, A (2018)	A Case of Asymptomatic Large Aortopulmonary Window in an Adult: Role of Cardiac CT, CMRI, and 3D Printing Technology	1. Surgical Planning 2. Interventional Cardiology Planning	1. APW	1. To evaluate the anatomical defect and assess the procedural options using a patient-specific 3D model of APW.	No pre-defined outcome measures	1. 3D printed models can be used to determine the feasibility of device or surgical closure in a patient with APW. In this case the 3D printed model revealed that device closure is not feasible since the anatomical defect did not have an inner rim.

Olejnik, P (2017)	Utilization of Three-Dimensional Printed Heart Models for Operative Planning of Complex Congenital Heart Defects	1. Surgical Planning 2. Interventional Cardiology Planning	1. IAA type A and APW type 2 2. DORV and CoA 3. TOF with multiple aortopulmonary collaterals 4. TOF status post transannular patch	1. To demonstrate the accuracy of the 3D printed cardiac models. 2. To describe the impact of 3D printed models on operative planning.	1. Model size (mm) 2. Digital image size (mm) 3. Intracardiac size (mm)	1. Patient-specific cardiac models were helpful to appreciate 3D intracardiac anatomy and for surgical/ interventional cardiology decision making. 2. Models improved spatial orientation. 3. Models were accurate in size compared to in vivo structures and digital image.
Jaworski, R (2017)	Three-Dimensional Printing Technology Supports Surgery Planning in Patients with Complex Congenital Heart	1. Surgical Planning 2. Interventional Cardiology Planning	1. PA and VSD status post Blalock–Taussig shunt	1. To describe the utility of a 3D printed model in surgical planning in a patient with PA and VSD.	No pre-defined outcome measures	1. A patient-specific 3D printed model of PA and VSD successfully guided surgical and interventional planning
McGovern, E (2017)	Clinical Application of Three-Dimensional Printing to the Management of Complex Univentricular Hearts with Abnormal Systemic or Pulmonary Venous Drainage	1. Surgical Planning 2. Interventional Cardiology Planning	1. Left atrial isomerism, interruption of the inferior caval vein with azygous continuation to a single right superior caval vein, PAPVC, superior-inferior arrangement of the ventricles, AV and VA discordance status post Fontan procedure 2. Right atrial isomerism, Dextrocardia, bilateral superior caval veins, left ventricular dominant atrioventricular septal defect, TGA, and PS 3. HLHS, MS and AS, right-sided infra-diaphragmatic anomalous pulmonary drainage to the inferior caval vein status post Blalock–Taussig shunt shunt and Glenn	1. To assess the utility of three-dimensional printed cardiac models in assisting with preoperative planning for children with a univentricular heart and abnormal systemic or pulmonary venous drainage.	No pre-defined outcome measures	1. Patient-specific 3D printed models can guide preoperative planning for patients with univentricular heart physiology and abnormal systemic or pulmonary venous drainage. In this study, 3D model led to decision of medical management in 2/3 cases.

Hoashi, T (2018)	Utility of a Super-Flexible Three-Dimensional Printed Heart Model in Congenital Heart Surgery	1. Surgical Simulation	1. DORV 2. ccTGA 3. TGA with intramural coronary running 4. IAA Type B with LVOT 5. TOF 6. PA with MAPCA's 7. HLHS	1. To assess the utility of 3D models for pre-operative simulation.	1. Cardiopulmonary bypass time (mins) 2. Aortic cross clamp time (mins)	1. Patient-specific 3D printed heart models can assist with understanding the relationship between intraventricular communications and great vessels and with simulations for the creation of intracardiac pathways.
Chen, S (2018)	Digital Design and 3D Printing of Aortic Arch Reconstruction in HLHS for Surgical Simulation and Training	1. Surgical Simulation	1. HLHS	1. To create a surgical training model using a patient-specific 3D model for aortic arch reconstruction in HLHS.	No pre-defined outcome measures	1. Patient-specific 3D printed cardiac models can be used for surgical simulation of neo-aortic arch reconstruction in HLHS.
Shiraishi, I (2010)	Simulative Operation on Congenital Heart Disease Using Rubber-Like Urethane Stereolithographic Biomodels Based on 3D Datasets of Multislice Computed Tomography	1. Surgical Simulation	1. HLHS 2. IAA 3. PA with VSD 4. DORV 5. CoA 6. TAPVC 7. PDA	1. To use 3D printed cardiac models for anatomical diagnosis and surgical simulation of complex congenital heart lesions.	No pre-defined outcome measures	1. Patient specific epoxy or rubber-like urethane 3D cardiac models accurately depict the anatomy of complicated CHD lesions and can assist with accurate diagnosis of the lesions. 2. The rubber-like urethane models can be used in preoperative surgical simulation.
Ilina, A (2017)	Patient-Specific Pediatric Silicone Heart Valve Models Based on 3D Ultrasound	1. Surgical Simulation	1. HLHS	1. To create silicone molded heart valve models from 3D printed heart valve molds. 2. To simulate surgery and suturing technique on 3D heart valve model.	1. Cardiac surgeons and fellows' feedback on usefulness of 3D printed heart valve models in training and surgery planning as measured by a questionnaire	1. 3D valve models created from 3D printed molds can be used in medical training and to practice or simulate surgery.
Amdani, S (2018)	Transcatheter Therapy of Anomalous Systemic Venous Drainage	1. Interventional Cardiology Planning	1. Anomalous drainage of right superior caval vein to the left atrium	1. To describe the use of a 3D printed cardiac model to plan a cardiac catheterization to occlude an anomalous right superior vena cava drainage to the left atrium.	No pre-defined outcome measures	1. A patient-specific 3D cardiac models assisted with procedural planning to correct anomalous systemic venous drainage of right superior caval vein to left atrium via transcatheter approach.

Lodzinski, P (2017)	Three-Dimensional Print Facilitated Ventricular Tachycardia Ablation in Patient with Corrected Congenital Heart Disease	1. Interventional Cardiology Planning	1. TOF with PA status post repair	1. To report the use of a 3D printed cardiac model to plan a cardiac catheter ablation procedure of ventricular tachycardia in a patient with TOF.	No pre-defined outcome measures	1. A patient-specific 3D printed model facilitated the ablation procedure in a patient with corrected TOF.
Yang, F (2015)	A Case of Transcatheter Closure of Inferior Vena Cava Type Atrial Septal Defect with Patent Ductus Arteriosus Occlusion Device Guided by 3D Printing Technology	1. Interventional Cardiology Planning	1. ASD	1. To report a case in which 3D printing technology was used to aid the treatment decision making for a boy with ASD.	1. Selection of the appropriate sized plug for ASD occlusion	1. Patient-specific 3D printed cardiac model assisted with utilizing PD plugs for ASD occlusion by delineating the patient's anatomy in detail.
Schievano, S (2007)	Percutaneous Pulmonary Valve Implantation Based on Rapid Prototyping of Right Ventricular Outflow Tract and Pulmonary Trunk from MR Data	1. Interventional Cardiology Planning	1. TOF	1. To compare the use of MRI to a 3D printed model for correct selection of percutaneous pulmonary valve implantation, in comparison to retrospective cases.	1. The success rate of percutaneous pulmonary valve implantation	1. Patient-specific 3D printed models allow more accurate patient selection for percutaneous pulmonary valve implantation compared to MRI data set alone.
Zhu, Y (2017)	Initial Study of Transthoracic Echocardiography Guided Three-Dimensional Printing on the Application of Assessment of Structural Heart Disease	1. Diagnostic	1. Endocardial cushion defect 2. ASD 3. MS 4. TOF 5. VSD	1. To investigate diagnostic value of 3D printed cardiac models.	1. Correct diagnosis of lesion with 3D model or 3D transthoracic echo compared to gold standard (intraopertative findings)	1. Patient-specific 3D cardiac models provide essential information for preoperative evaluation but diagnosis using 3D models was not statistically different than the gold standard (intraoperative diagnosis).

Garekar, S (2016)	Clinical Application and Multidisciplinary Assessment of Three Dimensional Printing in Double Outlet Right Ventricle With Remote Ventricular Septal Defect	1. Diagnostic	1. DORV 2. VSD	1. To assess the feasibility of using patient specific 3D printed model for diagnosis and clinical decision making.	1. Accuracy of Spatial orientation 2. Accuracy of relationship between aorta and the main pulmonary artery 3. Accuracy of relationship between aortic and pulmonary valve 4. Accuracy of VSD size 5. Accuracy of VSD position 6. Accuracy of Predicted baffle length 7. Accuracy of Tricuspid valve chordae 8. Accuracy of Aortic valve 9. Accuracy of RVOT 10. Accuracy of predicted RVOT obstruction by baffle 11. Accuracy of Predicted RV encroachment by baffle 12. Accuracy of Decision-making	1. Patient-specific 3D printed heart models are useful in pre-surgical planning in complex DORV 2. 3D printed models facilitate multidisciplinary team involvement and may help to mitigate communication gaps between the cardiologist/ radiologist and the surgeon. 3. The preoperative confidence level in contemplating, choosing, and carrying out a biventricular repair was enhanced by the opportunity to study life-size 3D models derived from patient-specific data in three patients
Vranicar, M (2008)	The Use of Stereolithographic Hand Held Models for Evaluation of Congenital Anomalies of the Great Arteries	1. Diagnostic	1. CoA 2. Vascular ring (not specified)	1. To retrospectively compare 3D printed cardiac models with procedural findings.	No pre-defined outcome measures	1. Patient-specific 3D printed cardiac models accurately displayed great artery anomalies.

Bhatla, P (2017)	Surgical Planning for a Complex Double-Outlet Right Ventricle Using 3D Printing	1. Surgical Planning 2. Surgical Simulation	1. DORV	1. To describe a case where a 3D printed cardiac model was used to simulate and plan surgical correction of DORV.	No pre-defined outcome measures	1. A patient-specific 3D model of DORV assisted with simulation provided better understanding of the anatomy and guided decision regarding the surgical approach
Shirakawa, T (2016)	Morphological Analysis and Preoperative Simulation of a Double-Chambered Right Ventricle Using 3-Dimnesional Printing Technology	1. Surgical Planning 2. Surgical Simulation	1. Double chambered right ventricle	1. To describe a case of double chambered right ventricle where 3D printed model was used to plan and simulate surgery.	No pre-defined outcome measures	1. A patient-specific 3D cardiac model of double chambered right ventricle assisted with simulating a surgical view through an incision of the lesion 2. This model was used in surgical planning and decision making regarding the operative approach.
Kiraly, L (2016)	Three-Dimensional Printed Prototypes Refine the Anatomy of Post-Modified Norwood-1 Complex Aortic Arch Obstruction and Allow Presurgical Simulation of the Repair	1. Surgical Planning 2. Surgical Simulation	1. HLHS 2. AS and MS	1. To describe a case of HLHS where 2 patient-specific 3D models were made to assist in surgical planning and simulate surgical approach.	No pre-defined outcome measures	1. Patient-specific solid blood-pool and flexible 3D models were successfully used for surgical simulation and guidance of spatial orientation regarding aortic arch.
Medero, R (2017)	Patient-Specific in Vitro Models for Hemodynamic Analysis of Congenital Heart Disease: Additive Manufacturing Approach	1. Hemodynamic Evaluation	1. Total cavopulmonary connection	1. To use 3D printed models to measure hemodynamic velocity in vitro and compare to in vivo measurements.	1. Hemodynamic velocity (m/s) 2. Vorticity (1/s) 3. Kinetic energy (mJ) [measured using 4D flow MRI]	1. 3D printing technology can be used to create in vitro models of the cardiovascular system that can help to understand physiological flows and potentially predict surgical outcomes.
Roldan-Alzate, A (2014)	Hemodynamic Study of TCPC Using in vivo and in vitro 4D Flow MRI and Numerical Simulation	1. Hemodynamic Evaluation	1. Extracardiac total cavo-pulmonary connection 2. Atriopulmonary total cavo-pulmonary connection	1. To assess total cavopulmonary hemodynamics using 4D Flow MRI in patients (in vivo), in patient-specific physical (in vitro) models and computational fluid dynamic simulations.	1. Flow in SVC, IVC, and pulmonary arteries (L/min) 2. Flow ratio of pulmonary arteries	1. 4D flow MRI of perfused 3D printed cardiac models can be used to create, verify and fine tune computational fluid dynamic (CFD) models of total cavopulmonary connection anatomy that correlate with in vivo 4D flow MRI.

Biglino, G (2017)	Piloting the Use of Patient-Specific Cardiac Models as a Novel Tool to Facilitate Communication During Clinical Consultations	1. Patient and Family Education	1. TOF 2. TGA 3. CoA 4. PA 5. DORV 6. Ebstein anomaly	1. To assess the impact of providing a patient-specific 3D printed cardiac model of their CHD to patients during a transition clinic (from pediatric to adult care).	1. The patients' rating of health status and confidence in explaining their condition to others 2. Name and features of their CHD (as a surrogate for CHD knowledge) 3. impact of CHD on their lifestyle 4. Satisfaction with previous/current visits 5. Positive/negative features of the 3D model	1. Patient-specific 3D printed models improved patients' satisfaction with clinic visit, confidence in explaining and knowledge of their condition. 2. The model made 32% of participants anxious. 3. The models helped to facilitate clinic visit.

Biglino, G (2015)	3D-Manufactured Patient-Specific Models of Congenital Heart Defects for Communication in Clinical Practice: Feasibility and Acceptability	1. Patient and Family Education	1. CoA 2. PA, Fontan circulation 3. TOF 4. TGA 5. AS 6. BAV 7. VSD 8. ASD 9. ALCAPA 10. TAPVC 11. IAA 12. DORV 13. PDA with left SVC	1. To determine if the use of 3D printed cardiac models during clinical consultation can improve communication between health care providers and patients and their parents.	1. Satisfaction with the 3D model, its usefulness, and clarity of the explanation as measured by a rating scale (1-10) 2. Parental understanding of their child's CHD and their engagement according to the cardiologist as measured by a rating scale (1-10) 3. Parental knowledge as assessed by asking them to mark diagrams, tick keywords and provide free text answers. 4. The duration of consultations	1. Parents and cardiologists both found the 3D models to be very useful and helpful in engaging the parents in discussing CHDs. 2. Parental knowledge was not associated with their level of education and did not improve following their visit. 3. Consultations involving 3D models lasted on average 5 min longer.
Kiraly, L (2019)	Three-Dimensional Virtual and Printed Models Improve Preoperative Planning and Promote Patient-Safety in Complex Congenital and Pediatric Cardiac Surgery	1. Surgical Planning 2. Diagnostic	1. HLHS 2. Tricuspid 3. atresia 4. TOF 5. DORV	1. To describe the use of patient-specific 3D printed cardiac models for accurate diagnosis and surgical planning.	No pre-defined outcome measures	1. Patient-specific 3D printed models refined the diagnoses by providing additional anatomical information and contributed to surgical planning. 2. AV valves were not clearly delineated in the models.

Ngan, E (2006)	The Rapid Prototyping of Anatomic Models in Pulmonary Atresia.	1. Surgical Planning 2. Diagnostic	1. PA with VSD and MAPCA's	1. To assess the utility and accuracy of solid anatomic models constructed with rapid prototyping technology for surgical planning in patients with PA, VSD and MAPCAs.	1. The accuracy and utility of the 3D model assessed by a post-operative questionnaire filled by a cardiac surgeon 2. The accuracy of the 3D model assessed by a post-operative questionnaire by a cardiac radiologist	1. Patient-specific 3D models assisted with operative planning and accurately depicted MAPCAs compared to angiogram and surgery.
Bartel, T (2016)	Three-Dimensional Printing for Quality Management in Device Closure of Interatrial Communications	1. Diagnostic	1. Secundum ASD	1. To assess the feasibility of patient-specific 3D printed secundum ASD model to confirm adequate device placement post procedurally.	No pre-defined outcome measure	1. The post-procedural 3D printed cardiac model demonstrated ASD occluder device adequately filled the ASD and that venous inlets were not obstructed .
Biglino, G (2017)	The Perception of a Three-Dimensional-Printed Heart Model From the Perspective of Different Stakeholders: A Complex Case of Truncus Arteriosus	1. Medical Education 2. Patient and Family Education 3. Surgical Planning	1. Right aortic arch 2. Repaired VSD 3. Repaired TA 4. Branch pulmonary artery stenosis	1. To assess the perspectives of various stakeholders regarding the utility of 3D models.	1. Qualitative reports and quotes from stakeholders	1. Parents felt that the models helped them understand the anatomy and need for the operation. 2. The surgeons felt the model helped with understanding the 3D relationship between the RPA and aorta
Biglino, G (2015)	Involving Patients, Families and Medical Staff in the Evaluation of 3D Printing Models of Congenital Heart Disease	1. Medical Education 2. Patient and Family Education	1. TGA 2. TOF 3. PA 4. CoA 5. HLHS status post Blalock Taussig shunt 6. HLHS status post Glenn	1. To develop a participatory approach to use 3D printed cardiac models with patients, parents, clinicians and nurses.	1. Patients' understanding of their own CHD.	1. Patients felt that the models were easier to understand than clinical imaging. 2. The models were found to be more helpful in life size (rather than enlarged) for patient education 3. Parents found a patient-specific model more helpful than a lesion specific model (complex CHD)

Yoo, S (2017)	Hands-on Surgical Training of Congenital Heart Surgery Using 3-Dimensional Print Models	1. Surgical Simulation 2. Medical Education	1. DORV with subaortic VSD 2. DORV with remote VSD 3. HLHS for Norwood 4. TOF 5. TGA 6. DORV with subpulmonary VSD	1. To use 3D printed cardiac models for hands-on surgical training workshops. 2. To evaluate users' responses to the models used for surgical training.	Surgeons' perceptions regarding the following as measured by a questionnaire: 1. Whether model provided necessary information on pathology 2. Quality of model for surgical simulation 3. Consistency and elasticity of model material is similar to human myocardium 4. Model material was acceptable for surgical simulation 5. Whether the surgical simulation workshop is helpful for improving surgical skills 6. Whether they would like to incorporate a similar workshop in training program	1. All responders found the course helpful in improving their surgical skills. 2. All responders would consider including such sessions in the training programs. 3. All found responders found that the models showed the necessary pathologic findings. 4. Most responders found that the consistency and elasticity of the model material were different from those of the human myocardium. 5. The responders thought that the quality of the models was acceptable or manageable for surgical practice. 6. The major weaknesses listed were related to the print material and poor representation of the cardiac valves.
Pluchinotta, F (2016)	Treatment of Right Ventricular Outflow Tract Dysfunction: A Multimodality Approach	1. Interventional Cardiology Simulation 2. Patient and Family Education	1. PA and VSD status post repair	1. To assess the usefulness of using patient-specific 3D printed model of PA to simulate and plan percutaneous pulmonary valve replacement.	No pre-defined outcome measures	1. Patient-specific 3D models provided useful information about the relationship between the main pulmonary artery and left coronary artery.

Bhatla, P (2017)	Altering Management Decisions with Gained Anatomical Insight From a 3D Printed Model of a Complex Ventricular Septal Defect	1. Surgical Planning 2. Diagnostic	1. VSD	1. To describe the use of a 3D printed heart model to clarify morphology of an unusual inter-ventricular communication.	No pre-defined outcome measures	1. A patient-patient-specific 3D model guided the sonographer to optimize angle of spectral Doppler interrogation during in performing an echocardiogram. 2. Medical management was decided over surgical management
Deferm, S (2016)	3D-Printing in Congenital Cardiology: From Flatland to Spaceland	1. Surgical Planning 2. Patient and Family Education	1. TOF with PA and MAPCA's	1. To describe the use of a 3D printed model in surgical planning and to educate the patient about the procedure.	No pre-defined outcome measures	1. Patient specific 3D printed cardiac models were successfully produced pre and post-procedure. Pre-procedure model was used to plan surgery and post-procedure model was used to evaluate surgical results.

*AI= Aortic insufficiency; ALCAPA= Anomalous left coronary artery arising from the pulmonary artery; APW= Aortopulmonary window; AS= Aortic stenosis; ASD= Atrial septal defect; AV= atrioventricular; BAV= Bicuspid aortic valve; ccTGA= Congenitally corrected transposition of the great arteries; CHD= Congenital heart disease; CoA= Coarctation of the aorta; DOLV= Double outlet left ventricle; HLHS= Hypoplastic left heart syndrome; IAA= Interrupted aortic arch; IVC= Inferior vena cava; LVOT= Left ventricular outflow tract; MAPCA= Major aortopulmonary collateral arteries; MS= Mitral stenosis; PA= Pulmonary atresia; PAPVC= Partial anomalous pulmonary venous connection; PDA= Patent ductus arteriosus; RVOT= Right ventricular outflow tract; SVC= Superior vena cava; TA= Truncus arteriosus; TAPVC= Total anomalous pulmonary venous connection; TGA= Transposition of the great arteries; TOF= Tetralogy of Fallot; VA= Ventriculoarterial; VSD= Ventricular septal defect

Appendix C

Reference List of Included Studies in Scoping Review

1. Sahayaraj RA, Ramanan S, Subramanyan R, *et al.* 3D Printing to Model Surgical Repair of Complex Congenitally Corrected Transposition of the Great Arteries. *World J Pediatr Congenit Heart Surg* 2019;**10**:373–5.
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Appendix D

<u>Pediatric Cardiology MedEd Workshop Assessment</u>

Introduction: Please complete the questionnaire below and circle your response.

All responses will be kept anonymous.

How would you rate your knowledge about:

	Very Low	Low	Moderate	High	Very High
The Normal Heart?	1	2	3	4	5
Tetralogy of Fallot?	1	2	3	4	5
D-Transposition of the Great Arteries?	1	2	3	4	5

1) Very low; do not have knowledge about this topic
2) Low; have little knowledge about this topic
3) Moderate; have some knowledge about this topic but there is still more to learn
4) High; have good knowledge about this topic but there is still more to learn
5) Very high; have very good knowledge and know almost everything about this topic

1. What are the four (4) components of Tetralogy of Fallot?
    a) overriding aorta, ventricular septal defect, right ventricular hypertrophy, pulmonary stenosis
    b) atrial septal defect, pulmonary stenosis, coarctation of the aorta, single ventricle
    c) double outlet right ventricle, aortic stenosis, coarctation of the aorta, ventricular septal defect
    d) overriding aorta, atrial septal defect, left ventricular hypertrophy, pulmonary stenosis

2. For normal heart anatomy, which is the correct statement regarding the atrioventricular and great vessel relationships?
    a) AV concordance, VA concordance, Aorta is anterior, Pulmonary artery is posterior and rightward
    b) AV concordance, VA concordance, Aorta is posterior, Pulmonary artery is anterior and leftward
    c) AV concordance, VA discordance, Aorta is posterior, Pulmonary artery is anterior rightward
    d) AV concordance, VA discordance, Aorta is anterior, Pulmonary artery is posterior leftward

3. For tetralogy of Fallot, which is the correct statement regarding the atrioventricular and great vessel relationships?
    a) AV concordance, VA concordance, Aorta is anterior, Pulmonary artery is posterior and rightward
    b) AV concordance, VA concordance, Aorta is posterior, Pulmonary artery is anterior and leftward
    c) AV concordance, VA discordance, Aorta is posterior, Pulmonary artery is anterior rightward
    d) AV concordance, VA discordance, Aorta is anterior, Pulmonary artery is posterior leftward

4. For d-Transposition of the Great Arteries, which is the correct statement regarding the atrioventricular and great vessel relationships?
    a) AV concordance, VA concordance, Aorta is anterior and leftward, Pulmonary artery is rightward, right aortic arch
    b) AV concordance, VA concordance, Aorta is posterior and rightward, Pulmonary artery is leftward, right aortic arch
    c) AV concordance, VA discordance, Aorta is posterior and leftward, Pulmonary artery is rightward, left aortic arch
    d) AV concordance, VA discordance, Aorta is anterior and rightward, Pulmonary artery is posterior, left aortic arch

5. Which two anatomical conditions must be present for an overriding aorta to exist?
    a) Atrial septal defect & aorta directed to right ventricle
    b) Ventricular septal defect & aortic valve directed towards the right and left ventricle
    c) Apical ventricular septal defect & coarctation of the aorta
    d) Midmuscular ventricular septal defect & atrial septal defect

6. What 2 factors contribute to cyanosis in Tetralogy of Fallot?
    i. Right ventricular outflow tract obstruction
    ii. Small right ventricle
    iii. Ventricular septal defect
    iv. Aortic arch obstruction
    a) i and iv
    b) ii and iii
    c) i and iii
    d) ii and iv

7. When comparing the anatomy of Tetralogy of Fallot and Transposition of the Great Arteries, which of the following statements is/are correct:
    i. Both have the same great vessel orientation.
    ii. Only one has an obstructed great vessel outlet.
    iii. Only one has AV concordance and VA discordance.
    iv. The anatomy of the pulmonary valve explains the clinical saturation
    a) i, iii, iv
    b) ii, iii, iv
    c) i, iv
    d) ii, iii

8. In patients with Transposition of the Great Arteries, why is a balloon atrial septostomy performed?
    a) to decompress left atrial pressure
    b) to allow mixing of oxygenated and deoxygenated blood
    c) to create an atrial septal defect
    d) b and c

9. Which is true about the Blalock-Taussig (BT) Shunt?

a) It is a procedure to augment pulmonary blood flow
b) It is a procedure to palliate Transposition of the Great Arteries
c) It is a procedure to create a connection between the right subclavian artery and the right atrium
d) It is an interventional cardiac catheterization procedure

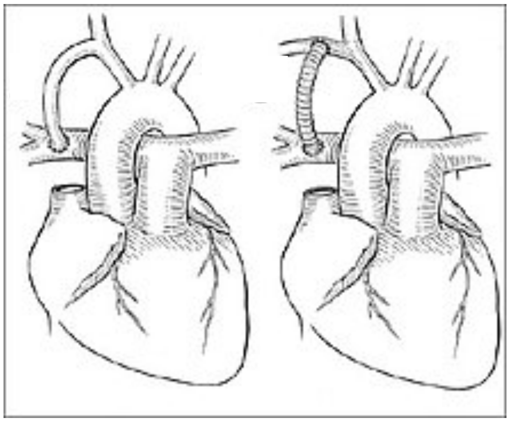
10. Which is true about d-Transposition of the Great Arteries?
    a) The great vessel have normal three dimensional relationship
    b) The right ventricle is hypertrophied
    c) Pulmonary and systemic blood flow are parallel circulations
    d) a and c

11. Match the cardiac chamber to great vessel relationships with the correct anatomy:
    i. Right Atrium to Left Ventricle to Pulmonary Artery/ Left Atrium to Right Ventricle to Aorta
    ii. Right Atrium to Left Ventricle to Aorta/ Left Atrium to Right Ventricle to Pulmonary Artery
    iii. Right Atrium to Right Ventricle to Pulmonary Artery/ Left Atrium to Left Ventricle to Aorta
    iv. Right Atrium to Right Ventricle to Aorta/ Left Atrium to Left Ventricle to Pulmonary Artery

    a) Normal Heart ______________
    b) Tetralogy of Fallot ______________
    c) D-Transposition of the Great Arteries ______________

12. For the following four (4) illustrations, name:
    1) the surgical procedure
    2) the most common underlying anatomic lesion for which it is used:

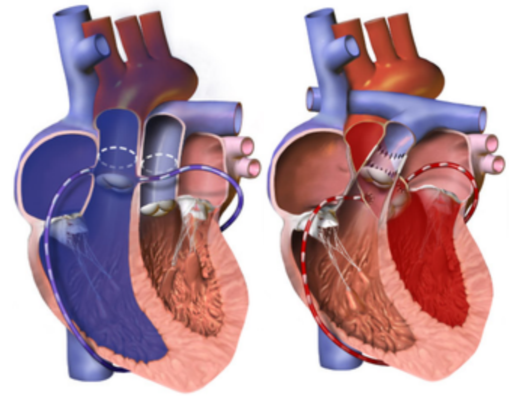
i.



1) ____________________

2) ____________________

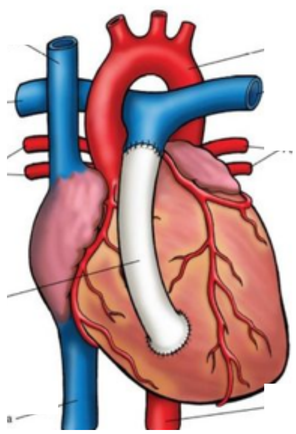
iii.



1) ____________________

2) ____________________

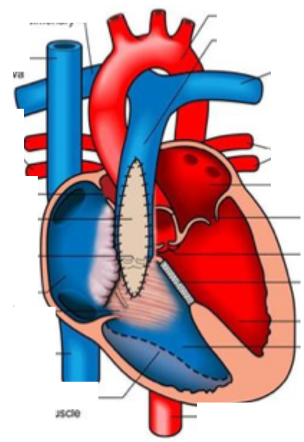
ii.



1) ____________________

2) ____________________

iv.



1) ____________________

2) ____________________

13. Jacob is a newborn patient with d-Transposition of the Great arteries and an intact ventricular septum. Which statement is incorrect about Jacob's condition?

a) Jacob's pulmonary and systemic circulations are in parallel
b) Blood from Jacob's right ventricle flows through his right ventricular outflow tract to his aortic valve.
c) Jacob will require surgical palliation within the first 6 weeks of life.
d) Jacob will be cyanotic at presentation.

14. Select the best answer to complete the following statement:
"The ___________is to d-Transposition of the Great Arteries as the_________ is to Tetralogy of Fallot."

a) Balloon Atrial Septostomy; BT Shunt
b) Arterial Switch; BT Shunt
c) Balloon Atrial Septostomy; Transannular Patch
d) Balloon Atrial Septostomy; RV to PA Conduit