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CASE REPORT

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Three-dimensional printing in a patient with pulmonary artery pseudoaneurysm and complex congenital heart disease—A case report

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Abstract

3D-printing is a powerful tool in patients with complex anatomy undergoing cardiac surgery.

KEYWORDS

3D printing, complex congenital heart disease, pediatric cardiac surgery, pulmonary artery pseudoaneurysm

1 | INTRODUCTION

Pulmonary artery pseudoaneurysms (PAPs) are dilatation of a focal pulmonary segment that may be congenital or acquired. Congenital PAP is commonly related to cardiovascular diseases, while acquired PAP is often a result of infection, trauma, or neoplasm.[1] The presentations of PAP vary and are dependent on the underlining etiology, location, and size. While hemoptysis is reportedly the most frequent presentation, other manifestations such as dyspnea and pneumonia are not uncommon.[2] Surgery is the treatment of choice when the pulmonary artery trunks are involved.

Computed tomography (CT) and magnetic resonance imaging (MRI) are imaging modalities traditionally used in patients with PAP. In patients who have complex anatomy, such as those with longstanding congenital heart diseases (CHD), these modalities may not provide enough intuitive spatial anatomy comprehension. 3D printing is a technique that creates a physical replica of the patient's anatomy, which enables detailed recognition of complex structures. In the last decade, 3D printing has been increasingly used in patients with complex anatomy and has proven itself a powerful tool in cardiovascular medicine.

In light of this, we present a case in which 3D printing was used in the perioperative management of a 23-year-old male with left PAP and complex CHD. Informed consent was given from the patient for publication.

2 | CASE REPORT

A 23-year-old male with a 3-year history of progressive dyspnea and hoarseness was referred from an outside clinic. Upon presentation, cyanosis, digital clubbing, and heart murmurs were noticed. Transthoracic echocardiogram (TTE) was ordered and revealed a common atrium, a single functional

right ventricle, double outlet right ventricle (DORV), and a patent ductus arteriosus (PDA). In addition to the findings on TTE, computed tomography angiography (CTA) also exposed a 63 mm PAP (Figure 1A, B). The PAP lies within the mediastinum, resulting in trachea compression. Multiple major aortopulmonary collateral arteries (MAPCA) between the pulmonary and systemic circulation were also reported. Incidentally, CTA (Figure 1B) also identified a midline liver, a right-sided stomach, two lobes in both lungs, and polysplenia, all of which are consistent with a diagnosis of heterotaxy syndrome. Subsequent cardiac catheterization also visualized two large MAPCA and determined that the patient had normal pulmonary vascular resistance (PVR). No other facial dysmorphism or cerebral anomaly was noted in this patient.

Considering the size of his pseudoaneurysm and its proximity to surrounding structures, a better appreciation of the complex anatomy is needed. Rapid prototyping improves both presurgical planning and intraoperative orientation in patients with complex anatomy,[3,4] we therefore took advantage of this technology for presurgical planning. Briefly, CTA data were imported into Mimics Innovation Suite 3D visualization software (Materialise Inc) for processing and segmentation to yield 3D-print ready stereolithography (STL) file. The STL file was subsequently exported and printed with plastic in color (HG Medical., J300 printer).

It can be appreciated from the 3D model (Figure 1C) that the upper end of the pseudoaneurysm reaches the level of the aortic arch, while its lower limit approximates the level of the carina. The large pseudoaneurysm compresses both the anterior trachea and main stem bronchus. Similar to the findings on CTA and catheterization, the 3D model also identified two MAPCA (Figure 1C, arrows): one bridges left subclavian artery and left pulmonary artery (PA) and the other connects the innominate artery and right PA. Oriented by the 3D model, we evacuated the hematoma and repaired a 50 mm tear in the pseudoaneurysm. A bidirectional Glenn procedure was also performed as palliation. The 3D model helped surgeons with presurgical planning and intraoperative orientation. The patient had an uneventful recovery.

3 | DISCUSSION

Pulmonary artery pseudoaneurysm is an uncommon but potentially lethal condition. It can be acquired or congenital, the latter of which is associated with CHD.[5-7] In CHD patients, pathological shunts between pulmonary and systemic circulatory systems can increase pulmonary blood flow and cause hypertension, both of which predispose to pulmonary artery dilation and pseudoaneurysm formation. The chief complaints in this patient are most likely results from pseudoaneurysm compression.

This patient had a delayed presentation of his CHD, one possible explanation is that PDA, MAPCA, and pulmonary

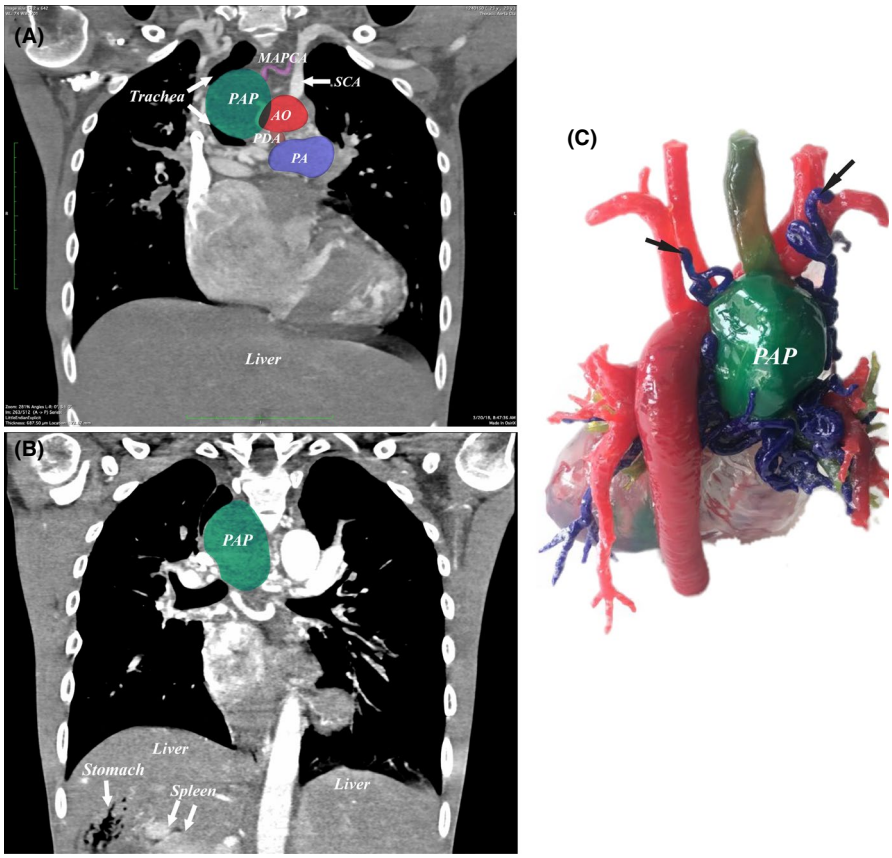

FIGURE 1 CT scans (coronal plane) and 3D model demonstrating patient anatomy. A, CT scan showing the relationship of PAP and surrounding structures. B, CT scan showing incidental findings in the patient consistent with heterotaxy syndrome. C, 3D printed model showing the relationship of the PAP with MAPCA (arrows) and surrounding tissue. AO, aorta; MAPCA, multiple major aortopulmonary collateral arteries; PA, pulmonary artery; PAP, pulmonary artery pseudoaneurysm; PDA, patent ductus arteriosus; SCA, subclavian artery

artery stenosis in this patient worked together as protection: The PDA and MAPCA provided adequate blood flow for tissue oxygenation, but the stenotic pulmonary artery also prevented the lungs vasculature from volume overload. Symptoms were delayed with adequate tissue oxygenation. Similarly, Chen et al[8] suggested that balanced pulmonary circulation is key to survival in uncorrected univentricular hearts.

In the current case, the pseudoaneurysm's size and proximity to nearby structures necessitate a deeper understanding of the spatial anatomy. Moreover, the collateral vessels in adults are fragile and prone to rupture during surgical manipulation. Ngan et al[9] reported that presurgical utilization of the 3D model was able to represent 96% of MAPCAs identified during surgery and 93% MAPCAs identified during angiography. We therefore employed 3D printing for presurgical planning. Oriented by the 3D model in the operating room, the MAPCA was identified and ligated without prolonged exploration and manipulation of tissue. Aside from the MAPCA, the 3D model also made it clear to the team that one can evacuate the hematoma from its superior end.

3D printing has been increasingly used in patients with complex anatomy in the last decade. It translates 2D images into a 3D physical replica and enables direct visualization of complex spatial anatomy. When used for presurgical planning, 3D printing can shorten exploration and decrease surgical time, both of which could help improve clinical outcomes.[3,10] Moreover, 3D printing can also improve the understanding of complex patient anatomy with presurgical stimulation. Costello et al[11] reported that stimulation with 3D models improved structure comprehension among residents. Yoo et al[12] recommended that trainees or inexperienced surgeons practice their skills on 3D-models before proceeding with specific operations. Finally, 3D printing can improve communication between patients and surgeons: Biglino et al[13] and Anwar et al[14] reported positive feedbacks from patients and their family members. In our own experience, we feel that 3D models improved presurgical planning, decreased surgery time and postoperative complications. 3D printing is not without limitations, cost is the major issue hampering its wide application. Additionally, there is no standard protocol from imaging to printing. The different acquisition and processing in imaging, segmentation techniques, and materials used in final printing can all affect the quality of the model and may eventually impact the quality of care. However, we also believe these limitations can all be overcome with the advancement of techniques over time.

4 | CONCLUSION

Through this report, we would like to emphasize on the advantages of 3D-printing in patients with complex anatomy. We expect it to open the door to personalized medicine in the future.

CONFLICT OF INTEREST

The authors here consent that this manuscript has not been previously published, nor is it under consideration in any other peer-reviewed media. All authors have significantly contributed to the treatment of the patient and the drafting of the manuscript. To the best of our knowledge, none of the authors have any conflict of interest, financial, and/or otherwise.

AUTHOR CONTRIBUTIONS

YZ: collected data and wrote the paper. XEZ: involved in image processing and wrote the paper. QL: initiated 3D printing strategy and performed surgery. HY: contributed resources and performed surgery.

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