JACC: CASE REPORTS VOL. 29, 2024

CONGENITAL HEART DISEASE

CASE REPORT: TECHNICAL CORNER

Immersive 3-Dimensional Visualization Aids Transcatheter Management of a Patient With Multiple Pulmonary Arteriovenous Malformations

Aishwarya Gurav, MD, DM,[a] Sarin Xavier, MTech,[b] Harikrishnan Anil Maya, MSc,[b] Rajiv Chandrasekharan, MD, DM,[c] Raman Krishna Kumar, MD, DM,[a] Mahesh Kappanayil, FNB[a]

ABSTRACT

Cutting-edge 3-dimensional technologies like 3-dimensional printing and extended reality visualization provide novel, immersive ways to understand and interact with volumetric medical imaging data for preprocedural planning. We present a case that illustrates the utility of these techniques in a patient requiring a complex transcatheter intervention. (JACC Case Rep. 2024;29:102480) © 2024 The Authors. Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 49-year-old man presented with gradually progressive exertional dyspnea and fatigue for 7 months (NYHA functional class II). Physical examination revealed pandigital clubbing, central cyanosis, and multiple telangiectasias. Room air oxygen saturation was 89% (Table 1).

PAST MEDICAL HISTORY

The patient gave a history of intermittent self-limiting episodes of epistaxis from 3 years of age (Table 1). A provisional diagnosis of hereditary

LEARNING OBJECTIVES

- To understand the importance of advanced 3D technologies in preplanning and executive complex vascular procedures.
- To select the most appropriate devices and sizes for planning closures of multiple and complex pulmonary arteriovenous malformations.
- To take advantage of the increasing levels of access to 3-dimensional technologies to generate personalized anatomical models that may change how we approach complex cases.

From the [a]Department of Pediatric Cardiology, Amrita Institute of Medical Sciences, Kochi, Kerala; [b]3D Printing and Extended Reality Lab, Amrita Institute of Medical Sciences, Kochi, Kerala; and the [c]Department of Adult Cardiology, Amrita Institute of Medical Sciences, Kochi, Kerala.

The authors attest they are in compliance with human studies committees and animal welfare regulations of the authors' institutions and Food and Drug Administration guidelines, including patient consent where appropriate. For more information, visit the Author Center.

Manuscript received April 1, 2024; revised manuscript received June 25, 2024, accepted June 28, 2024.

ISSN 2666-0849 https://doi.org/10.1016/j.jaccas.2024.102480

ABBREVIATIONS AND ACRONYMS

3D = 3-dimensional

AVM = arteriovenous malformation

VR = virtual reality

hemorrhagic telangiectasia was made based on the Curacao criteria.[1] As a known patient with type 2 diabetes mellitus, he was continued on sitagliptin 50-mg tablet once a day, dapagliflozin 10-mg tablet once a day, glimepiride-metformin combination 2-mg/500-mg tablet once a day, and atorvastatin 10-mg tablet once a day.

DIFFERENTIAL DIAGNOSIS

Respiratory disorders and Eisenmenger syndrome as alternative diagnoses were excluded at the index visit (**Table 2**).

INVESTIGATIONS

A chest x-ray showed homogeneous opacities in the right lung field. A contrast-enhanced computed tomography pulmonary angiogram acquired on Phillips multidetector (256-slice) scanner, with 0.8-mm slice thickness, showed 3 discrete pulmonary arteriovenous malformations (AVMs) in the right lung, 1 in the upper lobe and 2 in the lower lobe (**Figure 1**). A magnetic resonance angiogram of the brain excluded cerebral AVMs.

MANAGEMENT

Transcatheter occlusion of the pulmonary AVMs appeared to be technically feasible. However, significant procedural challenges were anticipated because of the multiplicity of the lesions and complexity of their locations within the pulmonary vascular tree, requiring multiple occlusion devices, precise wire-catheter manipulations, multiple intraprocedure angiograms, and significant radiation exposure. Although conventional workstation-based computed tomography visualization provided sufficient diagnostic information, it was insufficient for precise procedural planning. To overcome these challenges, advanced 3-dimensional (3D) visualization technologies were used.

Computed tomography DICOM (Digital Imaging and Communications in Medicine) data was segmented (Video 1) (by an experienced imaging expert) directly in virtual reality (VR) software Elucis (Realize Medical, Canada) and immersively visualized by the interventional team using an HTV Vive Pro VR headset (Video 2). Pulmonary vascular anatomy that was precisely segmented was visualized on a 1:1 scale, with hand-held controllers allowing manipulation in all planes, virtual dissections, measurements, and annotations (Video 3). The angulations and take-off of the lobar branches bearing the AVMs could be understood, and catheter approaches were planned. Sizes and deployment strategies for the occlusion devices were meticulously planned. Scaling-up of the virtual model also allowed virtual walk-throughs through the anatomy. The segmented 3D file (standard triangulation language format) was also converted into a 1:1 scale 3D-printed prototype using a fused deposition modeling printer (Ultimaker 3, Ultimaker B.V., The Netherlands) and polylactic acid material (**Figure 2**) to complement the VR visualization. Thus, whereas VR allowed the interventional team to create a mental roadmap, the 3D-printed model provided a physical guide for the procedure that could be carried to the catheterization laboratory. The entire process of conversion of the standard Digital Imaging and Communications in Medicine dataset to VR and 3D print was accomplished at the in-house point-of-care Pediatric Cardiac 3D Printing and Extended Reality Lab facility within our institution.

Through the 7-F right femoral venous access, a combination of a 7-F guiding catheter (Cook Medical) and a 0.035-inch Terumo wire was used to enter the right pulmonary artery. The pulmonary artery pressures were normal. A pulmonary angiogram was avoided. The 3D-printed model and the VR visualization were used as navigators and guiding tools to manipulate the catheter and wire combination for all 3 lesions. The Terumo wire was looped into the aneurysm (pulmonary AVM 1) and an angiogram performed to delineate the aneurysm (Video 4). The feeder artery of the upper branch of the right pulmonary artery measured around 5 mm, which correlated with that measured with the 3D model. A 10-mm Lifetech Cera vascular occluder was deployed in the distal-most part of the feeder artery, just at the entry site of the aneurysm, as per the plan (Video 5).

TABLE 1 Clinical Presentation and Basic Investigations of the Patient

Clinical characteristics	1. Gradually progressive exertional dyspnea (NYHA functional class II) 2. History of intermittent self-limiting episodes of epistaxis 3. Significant family history (history of similar episodes of epistaxis in first- and second-degree relatives)
Physical examination	1. Grade III pandigital clubbing 2. Central cyanosis 3. Multiple, small, red macular, telangiectasias on inner mucosa of lower lips 4. Pulse oximetry (Spo_2 = 89%) 5. Cardiovascular and respiratory system examinations were normal
Investigations	1. Arterial blood gas: Spo_2 = 91% preprocedurally and 97% postprocedurally 2. Chest x-ray: Homogenous opacities in the upper and lower zones of the right lung 3. Computed tomography showed 3 distinct lesions suggesting pulmonary arteriovenous malformations in the right lung field

TABLE 2 Approach to the Differential Diagnosis of Central Cyanosis

	Cyanotic Heart Defect	Pulmonary Diseases	PAVMs	Methemoglobinemia
Onset of cyanosis	Usually at birth or early childhood	Usually late after the development of respiratory symptoms	Any age	Congenital: from birth or infancy Acquired: presents at any age
Clubbing	Marked +++	++	+	Absent
Secondary erythrocytosis	Marked +++	+	+	Absent
Other features	Cyanotic spells, squatting, dyspnea on exertion, etc	Dyspnea, wheeze, etc	Usually cardiorespiratory symptoms absent	Cardiorespiratory symptoms absent; neurologic signs in type II congenital methemoglobinemia
Auscultation	Cardiac murmur present	Murmur usually absent; clinical respiratory signs present	Continuous bruit in large malformations; may be absent in diffuse small PAVMs	Normal
X-ray	Abnormal cardiac silhouette	Abnormal lung fields	Large AVMs: opacity in lung fields Diffuse small PAVMs: x-ray may be normal	Normal
Echocardiography	Abnormal heart	Usually normal; may show secondary pulmonary arterial hypertension	Contrast diagnostic echocardiogram: contrast appears in left-sided chambers after 3 to 4 cardiac cycles	Normal
Arterial blood gas	Spo_2 and Po_2 low	Spo_2 and Po_2 low	Spo_2 and Po_2 low	Spo_2 low, Po_2 normal

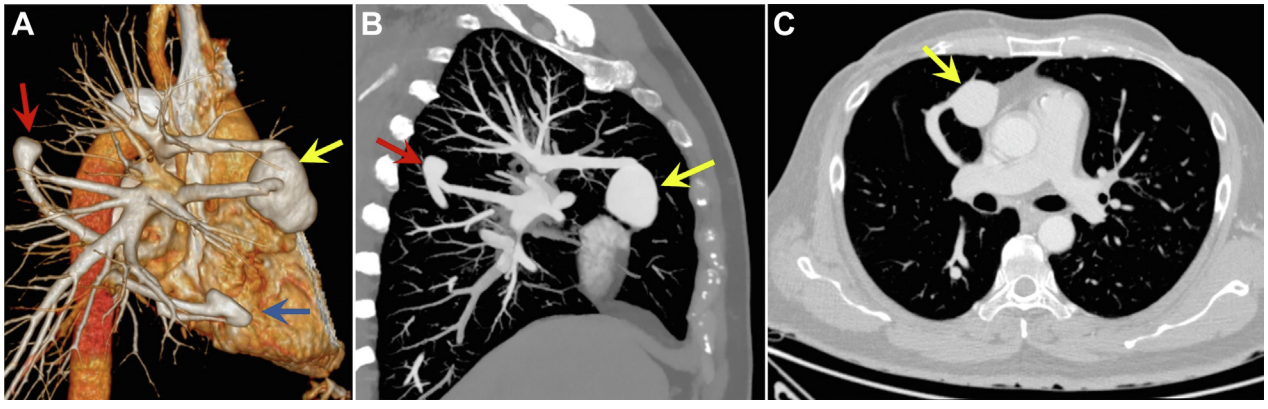
PAVM = pulmonary arteriovenous malformation.

For the second lesion, a combination of a 6-F guide catheter and a 0.035-inch Terumo wire was used to reach the distal-most part of the feeder branch of the right pulmonary artery supplying the second lesion, and angiograms were obtained to delineate the aneurysm. As per the prior plan, a 5 × 3 Lifetech Konar multifunctional ventricular septal defect occluder was deployed. Similarly, the third lesion was delineated and demarcated with the use of a 3D-printed model, and a 6-4 Konar multifunctional ventricular septal defect was used to occlude the aneurysm (Figure 3). Postprocedure saturations improved to 96% to 98% on room air. The total procedure time was 90 minutes, and fluoroscopy time was 35 minutes. The patient was started on aspirin 150 mg tablet once a day postprocedure and continued on it for 6 months.

DISCUSSION

Percutaneous transcatheter embolization has largely replaced surgical options as the mainstream treatment. A precise understanding of the complex angioarchitecture is of the utmost importance.

FIGURE 1 Computed Tomography Digital Imaging and Communications in Medicine (DICOM) Images in the Axial and Sagittal Planes Delineating Pulmonary AVMs



(A) Three-dimensional reconstruction from raw DICOM images of pulmonary AVM 1 (yellow arrow), pulmonary AVM 2 (red arrow), and pulmonary AVM 3 (blue arrow). (B) Raw DICOM image in the sagittal view. (C) Raw DICOM image in the axial view. AVM = arteriovenous malformation; DICOM = Digital Imaging and Communications in Medicine.

FIGURE 2 3-Dimensional Printed Model Showing the 3 Pulmonary Arteriovenous Malformations

Reports have described the use of 3D-printed models for planning treatment of cerebral AVMs,[2-4] aiding in the understanding of the angioarchitecture and preplanning procedure.

To the best of our knowledge, use of 3D technologies for transcatheter interventions for pulmonary AVMs has not been described. Immersive 3D visualization of the pulmonary vascular tree in our case enabled operators to gain excellent spatial understanding of the locations and morphology of the 3 discreet AVMs. Both 3D prints and VR visualizations, which were derived from computationally generated 3D files, allow immersive engagement, facilitating the spatial understanding of complex anatomies. Physical 3D prints provide excellent tactile engagement and easy manipulation, but the process is labor-intensive, requiring specialized hardware (3D printers), printing materials, printing time, and postprocessing.[5] Our 3D model required 8 hours, including time for printing and postprocessing. VR visualization, because it is purely digital, lacks tactile feedback but overcomes the challenges of creating physical prints, enabling scaling-up, virtual dissections, labeling, and measurements.[6,7]

In our case, we used both 3D print and VR visualization complementarily. The physical model enabled simulation with catheters and wires for an enhanced haptic understanding of the advances during the procedure, whereas VR allowed for virtual dissections, measurements, and preprocedural planning.

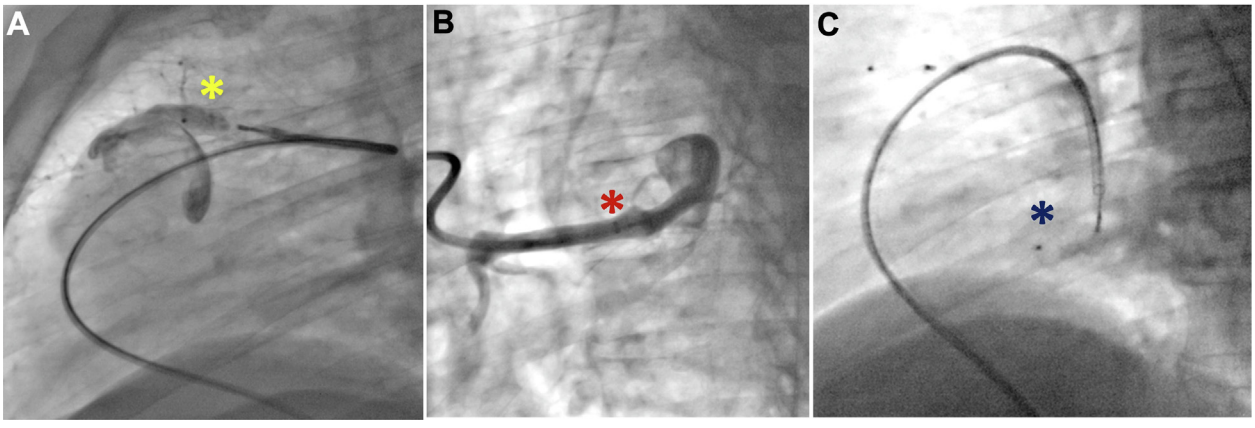
FOLLOW-UP

At 1 and 6 months of follow-up, the patient was in NYHA functional class I, and saturations were normal

CONCLUSIONS

Advanced 3D technologies can improve our ability to understand and treat complex vascular pathologies by helping operators to overcome the learning curve, shorten the duration of the procedure, and limit radiation exposure. These technologies will be increasingly used to perform patient-tailored complex vascular procedures.

FIGURE 3 Angiographic Images After Device Closure With the Device In Situ



(A) Yellow asterisk shows the 10- mm Lifetech Cera vascular occluder in situ sitting in the feeder artery of pulmonary arteriovenous malformation (AVM) 1. (B) Red asterisk shows the 5 × 3 Konar multifunctional ventricular septal defect (MF VSD) occluder in situ sitting in the feeder artery of pulmonary AVM 2. (C) Blue asterisk shows the 6-4 Konar MF VSD occluder in situ sitting in the feeder artery of pulmonary AVM 3.

FUNDING SUPPORT AND AUTHOR DISCLOSURES

The authors have reported that they have no relationships relevant to the contents of this paper to disclose.

ADDRESS FOR CORRESPONDENCE: Dr Mahesh Kappanayil, Department of Pediatric Cardiology, Amrita Institute of Medical Sciences and Research Centre, PO Ponekkara, Kochi, Kerala 68204, India. E-mail: maheshpeds@yahoo.co.in.

REFERENCES

1. Shovlin CL, Guttmacher AE, Buscarini E, et al. Diagnostic criteria for hereditary hemorrhagic telangiectasia (Rendu-Osler-Weber syndrome). *Am J Med Genet*. 2000;91(1):66-67.

2. Dong M, Chen G, Li J, et al. Three-dimensional brain arteriovenous malformation models for clinical use and resident training. *Medicine*. 2018;97(3):e9516.

3. Thawani JP, Pisapia JM, Singh N, et al. Three-dimensional printed modeling of an arteriovenous malformation including blood flow. *World Neurosurg*. 2016;90:675-683.

4. Chenoune Y, Tankyevych O, Li F, et al. Three-dimensional segmentation and symbolic representation of cerebral vessels on 3DRA images of arteriovenous malformations. *Comput Biol Med*. 2019;115:103489.

5. Averkin II, Grehov EV, Pervunina TM, et al. 3D-printing in preoperative planning in neonates with complex congenital heart defects. *J Matern Fetal Neonatal Med*. 2022;35(10):2020-2024.

6. Jung C, Wolff G, Wernly B, et al. Virtual and augmented reality in cardiovascular care: state-of-the-art and future perspectives. *Cardiovasc Imaging*. 2022;15(3):519-532.

7. Awori J, Friedman SD, Howard C, Kronmal R, Buddhe S. Comparative effectiveness of virtual reality (VR) vs 3D printed models of congenital heart disease in resident and nurse practitioner educational experience. *3D Print Med*. 2023;9(1):2.

KEY WORDS congenital heart defect, 3-dimensional imaging, 3-dimensional printing

APPENDIX For supplemental videos, please see the online version of this paper.