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CASE REPORT

ADVANCED

CLINICAL CASE

Printing the Procedure

Successful Closure of a Coronary Cameral Fistula With 3-Dimensional Model

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ABSTRACT

Cardiac 3-dimensional printing for pre-procedural planning of structural heart procedures is a promising new tool. Despite current potential drawbacks, 3-dimensional models can help cardiologists better understand and treat complex cardiac defects. We describe a successful coronary cameral fistula closure planned with the aid of a 3-dimensional model. (**Level of Difficulty: Advanced.**) (J Am Coll Cardiol Case Rep 2020;2:488–92) © 2020 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/).

PRESENTATION

A 29-year-old woman presented with complaints of fatigue, dyspnea on exertion, and palpitations. Her symptoms had worsened over the previous 6 months. She had a continuous murmur on the right sternal border. Her chest radiograph showed active congestion of the pulmonary vasculature. Her left ventricular size and diastolic and systolic functions were normal. The right heart chambers were slightly dilated with a pulmonary artery systolic pressure of 30 mm Hg. Turbulent flow in the right atrium was noted. Her cardiologist referred the patient with an initial diagnosis of atrial level left-to-right shunting.

LEARNING OBJECTIVES

- To understand the clinical approach for diagnosis and treatment of coronary fistulae.
- To understand the use of 3D printing for advanced structural intervention planning.

MEDICAL HISTORY

The patient had an uncomplicated pregnancy at 24 years of age. She had undergone a successful ablation procedure for atrioventricular nodal re-entrant tachycardia a year previously. After ablation, deep venous thrombosis developed, and she received 6-month anticoagulation therapy with rivaroxaban.

INVESTIGATIONS

Turbulent flow in the right atrium (RA) with a diastolic gradient of 60 mm Hg was noticed on transthoracic echocardiography. Transesophageal echocardiography was performed to determine the exact localization of intracardiac shunting. A coronary cameral fistula was diagnosed, and multidetector computed tomography (MDCT) was performed to better delineate the characteristics of the fistula. The fistula originated from the left main coronary artery (LMCA) and traveled posteriorly along a tortuous tract ending in the right atrium. The LMCA was dilated to a diameter of

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12.28 mm. The fistula's entrance at the LMCA was 9 mm, and the exit at the RA site was 15 mm. The narrowest point along the fistula's tract measured 11 mm (Figure 1).

DIFFERENTIAL DIAGNOSES

Right-sided volume overload, turbulent flow in the RA, and active congestion of the pulmonary vasculature without cyanosis raised consideration of atrium-level shunts including atrial septal defect, patent foramen ovale, Gerbode defect, partial anomalous pulmonary venous connection, and coronary cameral fistula.

MANAGEMENT

The size of the fistula was considered suitable for percutaneous closure. Tortuous tract and the high flow rate were among the factors favoring surgery. It was decided to plan the treatment with the help of a 3-dimensional (3D) printed model. The procedure's feasibility, device position, and stability and specific steps would first be analyzed by the simulation using a 3D model. If the procedural success was confirmed, percutaneous closure would be chosen over surgery.

The 3D printing of the model was performed in collaboration with an experienced laboratory. An intact hollow model of the fistula was designed from MDCT images. The 3D model was printed in 1:1 sizing of the fistula with the use of Mimics software (Materialise, Glen Burnie, Maryland) (Figure 2).

The possible closure site was determined as the narrowest point along the fistula tract. The appropriate occluder size would be 20% to 40% larger; thus, an Amplatzer 12-mm vascular plug (AVP) I (St. Jude Medical, St. Paul, Minnesota) was considered suitable. A substitute for the device was also printed (Figure 3). The substitute was transferred to the planned location on the 3D model by creating an arteriovenous (AV) loop, and the stability of the device was confirmed. After a simulation using the 3D model, the percutaneous closure proceeded.

The patient was transferred to the catheterization laboratory. Right femoral venous and arterial access was secured with 6-F sheaths (Terumo, Tokyo, Japan). The LMCA was cannulated with an Amplatz Left 2 guiding catheter (Medtronic, Manalapan, New Jersey). A Rubicon 35 5-F backup support catheter (Boston Scientific, Marlborough, Massachusetts) was advanced over the 0.014-inch Fielder wire (Asahi Kasei Corp., Tokyo, Japan). After the Rubicon catheter was positioned in the RA, the wire was exchanged with a hydrophilic 0.035-inch Radiofocus straight guidewire (Terumo). The AV loop was created by snaring the 0.035-inch wire in the pulmonary artery (Figure 4). A 7-F Destination Guiding sheath (Terumo) was advanced over the wire to the origin of the fistula in the RA for additional support. The AVP I was advanced anterogradely from the venous side through the preplanned position. After proper positioning of the AVP I, the operator ensured a stable device position with a wiggling maneuver, and the device was released. Complete closure of the fistula was confirmed without residual leakage by radiopaque injection from the LMCA (Figure 5). No complications were observed.

The patient was discharged uneventfully on the following day. Rivaroxaban treatment continued for 6 months.

ABBREVIATIONS AND ACRONYMS

AV = arteriovenous

LMCA = left main coronary artery

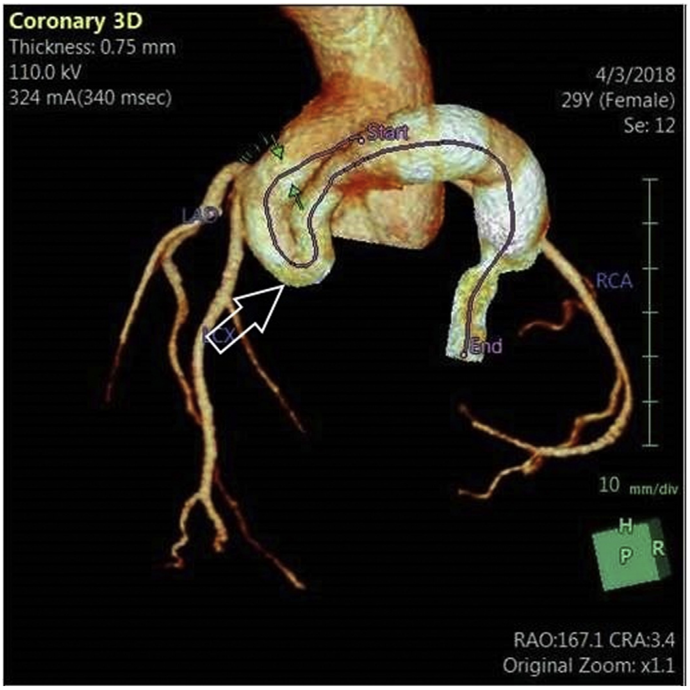
MDCT = multidetector-computed tomography

RA = right atrium

DISCUSSION

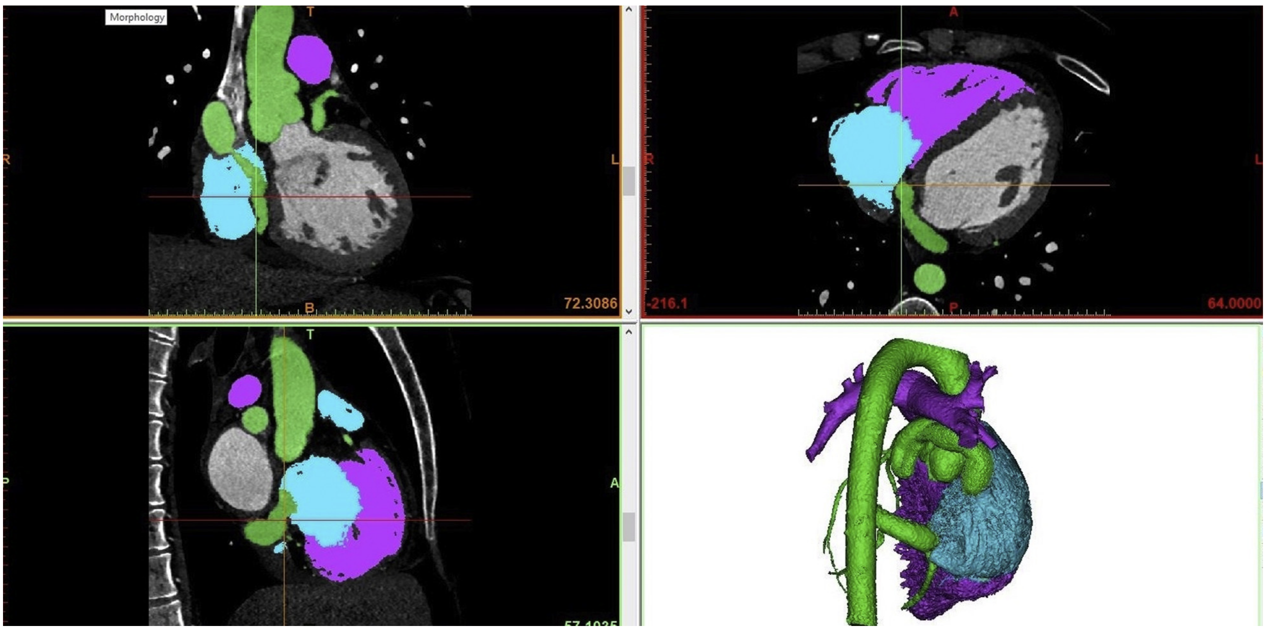
A coronary cameral fistula is a communication between 1 or more of the coronary arteries with a cardiac chamber (1). Surgery had been the treatment of choice for fistulae until Reidy et al. (2) described the first successful transcatheter closure. As the guidelines have no specific recommendations for choice of fistulae treatment procedures, the approach is

FIGURE 1 Reconstructed MDCT



Reconstructed MDCT shows the tortuous course and large diameter of a fistula's tract. The narrowest portion of the fistulous tract **(arrow)** is 11 mm. MDCT = multidetector-computed tomography.

FIGURE 2 **MDCT Images Processed With Mimics Software**

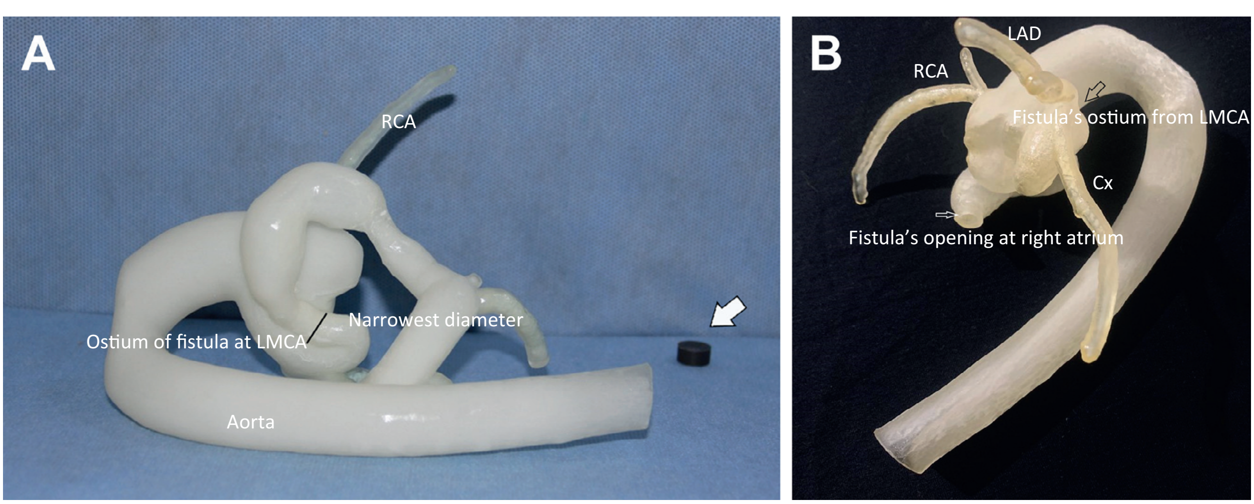


Two MDCT images processed with Mimics Software for 3-dimensional (3D) printing. Aorta, coronary arteries, and fistula are shown in **green**. Reconstructed image used for 3D printing is shown at the **bottom right**. MDCT = multidetector-computed tomography.

determined mainly by experience and on a case-by-case basis.

Device sizing and positioning are important, as embolization and the residual leakage would affect the result of a procedure of this complexity. Coils are used primarily in small to moderate fistulae, whereas for larger fistulae, Amplatzer devices are recommended (3,4). AVP I is retrievable as long as it is connected to the delivery cable, which is advantageous for interventions of this complexity.

FIGURE 3 **3-Dimensional-Printed Model of Fistula and Surrogate Closure Plug**



(A and B) 3-dimensional-printed model of the fistula and surrogate closure plug **(arrow)** used for planning the procedure. Although a 1:1 ratio was preserved for the fistula, the neighboring tissues were printed in a reduced size for the feasibility of simulation.

In the era of digitized medicine, cardiac 3D printing is valuable for diagnostic work-up, teaching purposes, tailoring treatment choices, simulating interventional procedures, and improving patient-physician communication (5). Oliveira-Santos et al. (6) reported a successful complex circumflex artery ostial stent restenosis treatment guided by simulation with a patient-specific 3D model. In congenital heart diseases, most reports used 3D printing for surgical planning with fewer examples of transcatheter procedures (7). There are even fewer reports of 3D printing for coronary fistulae treatment. Misra et al. (8) reported the use of a 3D printed model of a coronary-pulmonary artery fistula to guide surgical treatment. Forte et al. (9) reported experience with 3D printed models in coronary artery fistulae in 4 patients. The study concluded that 3D printing did not add much value for simple coronary artery fistulae diagnosis and treatment. In cases with complex anatomic features, 3D printing contributed to the interpretation of images and facilitated communication of morphology to the interventional team. In their series, only 1 simulation was successfully implemented in the real-life procedure. The limited ability of 3D printing to mimic tissue characteristics and flow properties might have restricted success of pre-procedural planning (5–7).

This study illustrated the feasibility and potential utility of 3D printing for planning transcatheter coronary cameral fistula closure. To the best of the present authors' knowledge, this is the first report of a 3D-printing-assisted procedural planning for a coronary fistula closure in a stepwise manner. The 3D model guided the device selection and procedural steps, enabling a successful procedure with decreased fluoroscopy time. The model enhanced communication about the procedure between team members and the patient and her family.

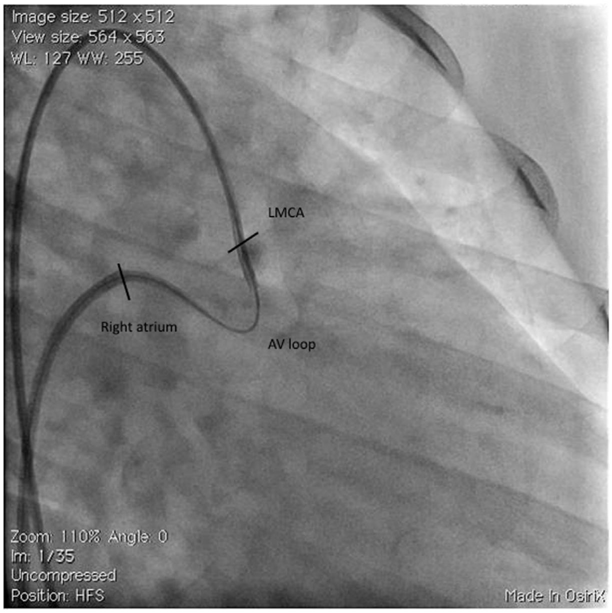
FOLLOW-UP

The patient was followed for the succeeding year. Her complaints disappeared, and she reported at her 12-month control visit the ability to exercise for 150 min per week. The right heart chamber sizes were normal, with pulmonary artery systolic pressure of 25 mm Hg. Transthoracic echocardiography showed no turbulent flow in the RA.

CONCLUSIONS

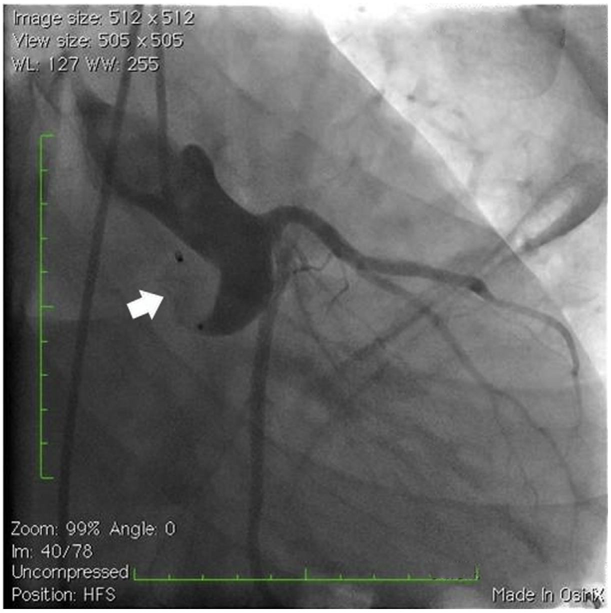
Cardiac 3D printing is a promising new tool for pre-procedural planning in transcatheter interventions for congenital heart diseases. There are still concerns of cost, model accuracy, limited ability to mimic tissue characteristics and flow properties, and lack of robust

FIGURE 4 LMCA Cannulated With an Amplatz Left 2 Guiding Catheter



The LMCA is shown cannulated with an Amplatz Left 2 guiding catheter. The AV loop was created, and a 7-F destination guiding sheath was advanced over the wire to the origin of the fistula in the right atrium. AV = atrioventricular; LMCA = left main coronary artery.

FIGURE 5 AVP I Is Placed at the Predetermined Landing Point



The AVP I **(arrow)** is placed at the pre-determined landing point anterogradely. The successful closure of the fistula was confirmed by radiopaque injection from the LMCA. AVP I = Amplatzer vascular plug I; LMCA = left main coronary artery.

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evidence before the widespread use of 3D printing in transcatheter procedures can be implemented.

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KEY WORDS 3-dimensional printing, coronary vessel anomaly, percutaneous coronary intervention