JACC: CASE REPORTS VOL. 4, NO. 18, 2022

CASE REPORT

INTERMEDIATE

CLINICAL CASE

Pulmonary Vein Occlusion After Surgical Atrial Fibrillation Ablation and Left Atrial Appendage Occlusion

Neel Patel, MD,[a] Moghniuddin Mohammed, MD,[a,b] Nachiket Apte, MD,[a] Sunthosh V. Parvathaneni, MD,[c] Seth Sheldon, MD,[a] Madhu Reddy, MD[a]

ABSTRACT

We report a case of pulmonary vein (PV) occlusion in a patient with a history of surgical atrial fibrillation ablation and left atrial appendage occlusion with unsuccessful endovascular management. Delayed diagnosis of PV stenosis post-ablation can make interventional treatment options less likely to be successful. (**Level of Difficulty: Intermediate.**)

HISTORY OF PRESENTATION

A 55-year-old man was referred for symptomatic left superior pulmonary vein (LSPV) occlusion management. His vital signs were within normal limits, and he was in sinus rhythm on presentation. Approximately 16 months earlier, he had a non–ST-segment elevation myocardial infarction and underwent coronary artery bypass grafting for severe coronary artery disease. Because of his history of paroxysmal atrial fibrillation (AF), he also underwent an ablation procedure using the Isolator Synergy Ablation System (AtriCure) and epicardial left atrial appendage (LAA) occlusion (LAAO) with AtriClip (AtriCure). The patient had left and right pulmonary vein (PV) isolation with clamps applied 3 times on each side with crossing lesions and was discharged with an amiodarone regimen.

LEARNING OBJECTIVES

- To recognize PVS as a possible complication of surgical AF.
- To recognize the importance of timely diagnosis and intervention of PVS in symptomatic patients to improve outcomes.

At 3-month follow-up, he had "asymptomatic" AF recurrences and was switched from amiodarone to sotalol. A subsequent transoesophageal echocardiogram (TEE) showed an occluded LAA with mildly increased LSPV flow velocity (1.5 m/s) with turbulence on color Doppler imaging.

At 9-month follow-up (7 months before presentation), he reported fatigue and dyspnea on exertion, which were suspected to be effects of sotalol, and the drug was discontinued. His symptoms did not improve, and the burden of AF increased, with incrementally worsening symptoms. Subsequently, he underwent catheter cryoballoon PV isolation (11-months after coronary artery bypass grafting with a maze procedure). During this procedure, he was found to have LSPV occlusion, which was confirmed by pulmonary venography and subsequent computed

From the [a]Department of Cardiovascular Medicine, The University of Kansas Health System, Kansas City, Kansas, USA; [b]Department of Biomedical and Health Informatics, University of Missouri Kansas City, Kansas City, Missouri, USA; and the [c]Department of Cardiology, Mercy Hospital, Springfield, Missouri, USA.
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 January 27, 2022; revised manuscript received June 22, 2022, accepted July 8, 2022.

ISSN 2666-0849 https://doi.org/10.1016/j.jaccas.2022.07.017

tomography angiogram (CTA). The patient was referred to our center (University of Kansas Health System, Kansas City, Kansas, USA) for further management.

PAST MEDICAL HISTORY

The patient's history included hypertension, dyslipidemia, obstructive sleep apnea, and paroxysmal AF.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included PV occlusion secondary to surgical AF ablation and/or extrinsic compression by an epicardial LAAO device.

INVESTIGATIONS

Ventilation-perfusion (V/Q) scan showed a significant mismatch with absent perfusion of the left upper lobe. The entire left lung contributed to only ~20% of total perfusion (Figure 1). A repeat CTA confirmed LSPV occlusion (Figures 2A, 2B, and 3A to 3D). The TEE showed an occluded LAA with a clip. The left inferior PV was patent with mild stenosis, and no flow was detected from the LSPV on color flow Doppler imaging (Figures 4A and 4B).

MANAGEMENT

After multidisciplinary team discussions, a transcatheter approach to relieving the occlusion with PV stenting was considered. There was an initial suspicion that the LAA clip was compressing and occluding the LSPV. However, it was ruled out on the basis of multimodality imaging because it was

ABBREVIATIONS AND ACRONYMS

AF = atrial fibrillation

CTA = computed tomography angiogram

LAA = left atrial appendage

LAAO = left atrial appendage occlusion

LSPV = left superior pulmonary vein

PV = pulmonary vein

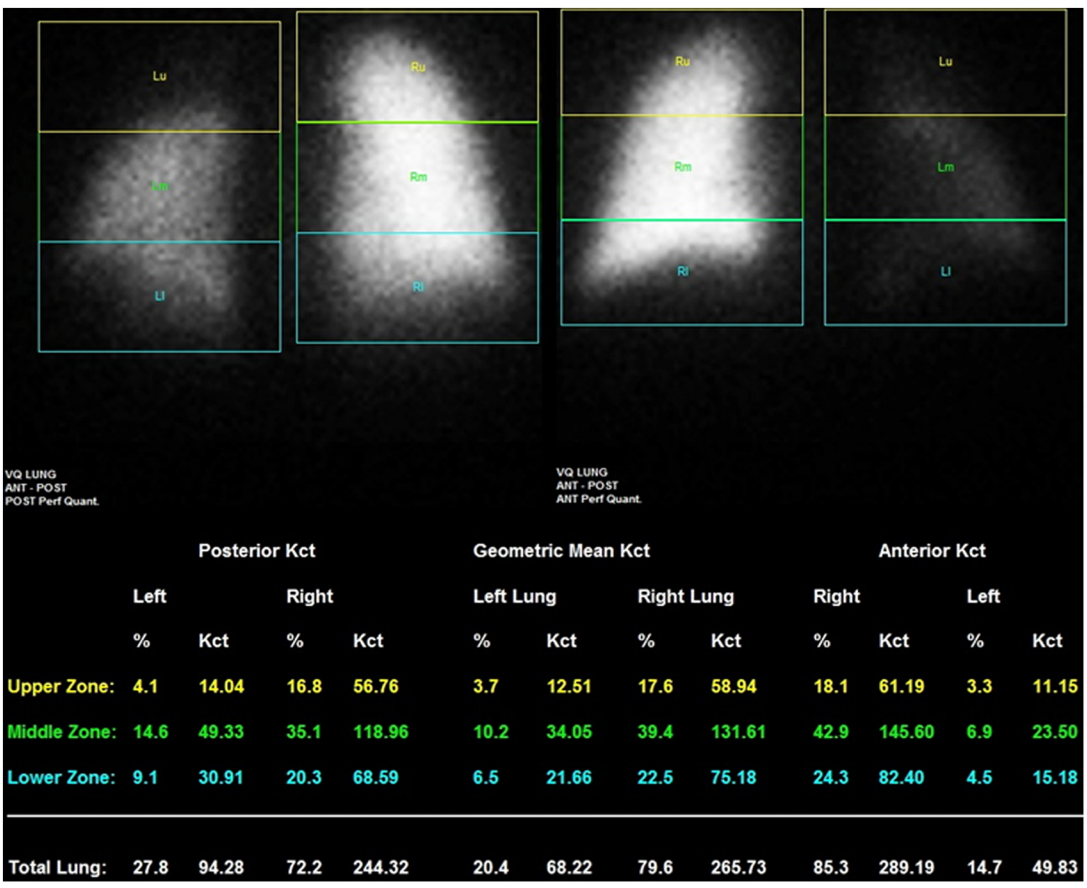
PVS = pulmonary vein stenosis

RF = radiofrequency

TEE = transesophageal echocardiogram

V/Q scan = ventilation-perfusion scan

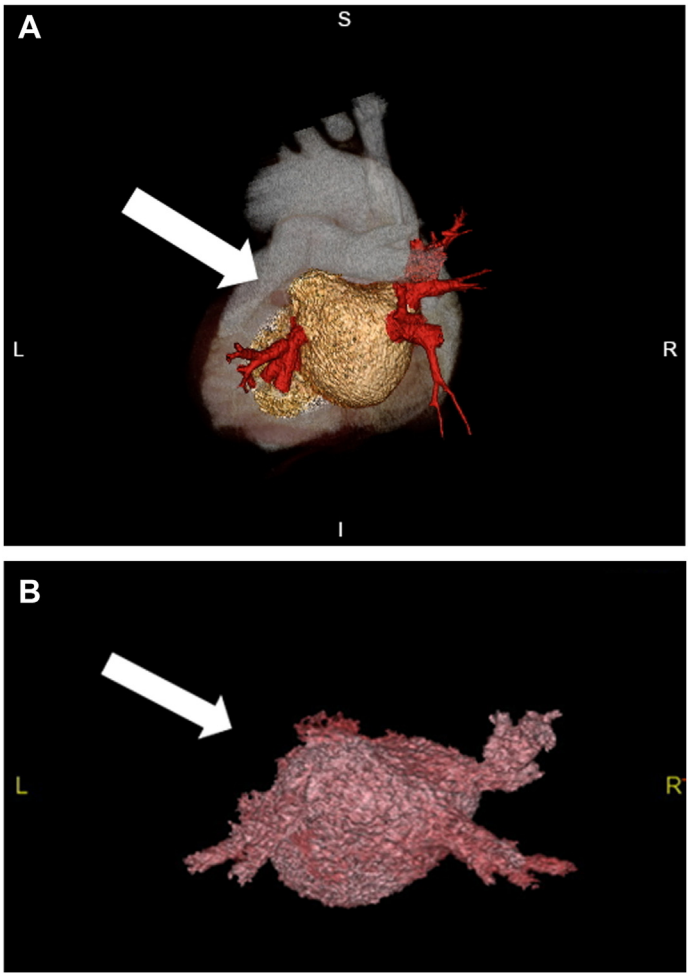
FIGURE 1 V/Q Scan Showing Decreased Perfusion of the Left Lung



	Posterior Kct				Geometric Mean Kct				Anterior Kct			
	Left		Right		Left Lung		Right Lung		Right		Left	
	%	Kct	%	Kct	%	Kct	%	Kct	%	Kct	%	Kct
Upper Zone:	4.1	14.04	16.8	56.76	3.7	12.51	17.6	58.94	18.1	61.19	3.3	11.15
Middle Zone:	14.6	49.33	35.1	118.96	10.2	34.05	39.4	131.61	42.9	145.60	6.9	23.50
Lower Zone:	9.1	30.91	20.3	68.59	6.5	21.66	22.5	75.18	24.3	82.40	4.5	15.18
Total Lung:	27.8	94.28	72.2	244.32	20.4	68.22	79.6	265.73	85.3	289.19	14.7	49.83

Absent perfusion involving the left upper lobe. The quantitative results obtained using a geometric mean for pulmonary perfusion showed the left lung supplying 20.4% of the total perfusion to the lungs. ANT-POST = anterior-posterior; Kct = kilocounts; POST Perf Quant = posterior quantitative perfusion analysis; V/Q = ventilation-perfusion.

FIGURE 2 Computed Tomography Angiography of the Heart



Imaging performed at **(A)** an outside hospital and **(B)** our institution (University of Kansas Health System, Kansas City, Kansas, USA) shows an occluded left superior pulmonary vein. The 3-dimensional reconstructions of the pulmonary veins show the occluded left superior pulmonary vein marked by **white arrows.** I = inferior; L = left; R = right; S = superior.

evident that the AtriClip was not compressing the LSPV (**Figures 5A to 5C**, Video 1). PV recanalization was attempted with multiple wires and catheters but was unsuccessful, and the procedure was aborted. The patient was discharged home on guideline-directed medical therapy for heart failure and was recommended the conventional measures for improving symptoms of PV stenosis (PVS), including weight loss and incentive spirometry to improve left lower and right lung capacity.

DISCUSSION

We report the first case of PV occlusion after surgical AF ablation using Isolator Synergy Ablation System. A synergy ablation system is a dual-electrode bipolar radiofrequency (RF) ablation device that creates pulsed RF energy between the 2 electrodes to create more uniform, column-shaped lesions.[1]

PVS remains a feared complication after endocardial catheter ablation for AF. The incidence has decreased from up to 3% in the past[2] to <1% at present because of improved techniques.[3] However, it is infrequent after surgical AF ablation. In our case, it was likely the result of distal ablation causing the transmural injury of the LSPV with a subsequent inflammatory response and progressive stenosis and occlusion. PVS is usually asymptomatic, but when it is symptomatic, the most common symptom is dyspnea[4]; others are cough, chest pain, fatigue, and hemoptysis.[5] Symptoms usually manifest 3 to 6 months after the ablation. Prompt identification and management are necessary to prevent progression to PV occlusion.

PVS can be diagnosed by multimodality imaging. CTA is the most commonly performed noninvasive test and provides a detailed analysis of the PV and surrounding structures. Magnetic resonance imaging can visualize PV and analyze flow dynamics by measuring velocity and gradients across the pulmonary arteries and veins while avoiding radiation to patients.[3] A V/Q scan can evaluate the functional significance of PVS but is less sensitive in mild or moderate stenosis, and other causes can influence its results. TEE has high sensitivity and specificity, 82% to 100% and 95% to 100%, respectively. The current guidelines provide information on diagnostic suspiciousness on the basis of symptoms. However, they do not include echocardiographic findings (ie, increased PV flow velocity >1.1 m/s combined with color Doppler turbulence, which was reported to be a reliable index[3,5]).

The management depends on the symptoms and severity of stenosis. Asymptomatic PVS from 50% to 80% can be monitored with imaging every 3 to 6 months. Some centers perform angioplasty for single PVS >75%, whereas others perform it for a

cumulative stenosis index of >75% on the ipsilateral side.[5] Delay in restoring patency may lead to progression of pathophysiologic vasoconstriction of the affected lung segments and gradual loss of perfusion. Moreover, there is a risk of rapid progression from PVS to total occlusion, thus making the intervention less likely to succeed.[2] The most common treatment is balloon angioplasty or stent placement. A recent meta-analysis comparing the 2 strategies showed stenting to be superior to isolated balloon angioplasty in maintaining patency and decreasing the need for reintervention.[6]

To our knowledge, there is only 1 reported case of symptomatic PVS after epicardial ablation that was successfully treated with balloon angioplasty.[7] Ours is the first case of PV occlusion after surgical AF ablation using the Synergy system. A challenging aspect of our case that could have contributed to an accelerated course and subsequent failure of transcatheter therapy is the presence of AtriClip anterior to the occlusion. Angiography showed that the extrinsic compression by AtriClip was less likely to have caused the PV occlusion. PVS with endocardial LAAO has been reported previously[8] but has not been reported as caused by epicardial LAAO with AtriClip. Previous studies and a more recent systematic review of AtriClip did not report any adverse events related to the device.[9] Further large trials evaluating AtriClip may provide more information if PVS can be caused by AtriClip alone.

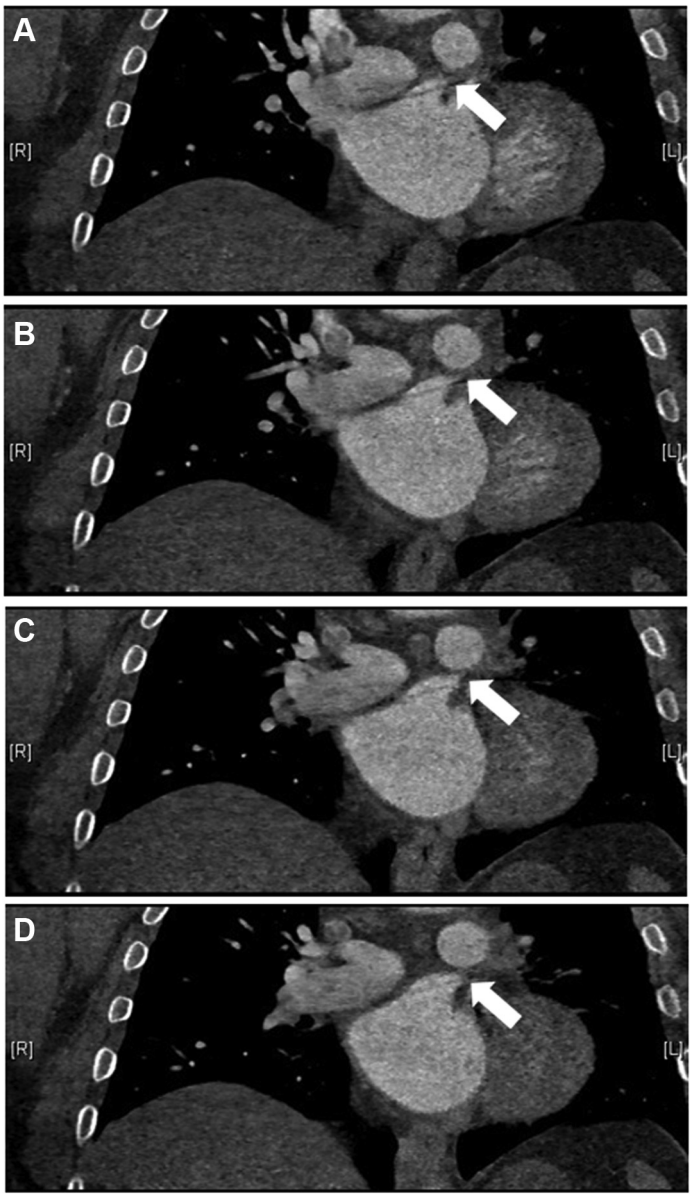
FOLLOW-UP

On 6-month follow-up, he continued to have dyspnea on exertion without significant progression of his symptoms or pulmonary pressures.

CONCLUSIONS

PVS is a possible complication after surgical AF ablation that requires a high level of suspicion. Prompt diagnosis and intervention are necessary to improve perfusion of the affected lung segments and prevent progression to total occlusion, which makes transcatheter therapy challenging.

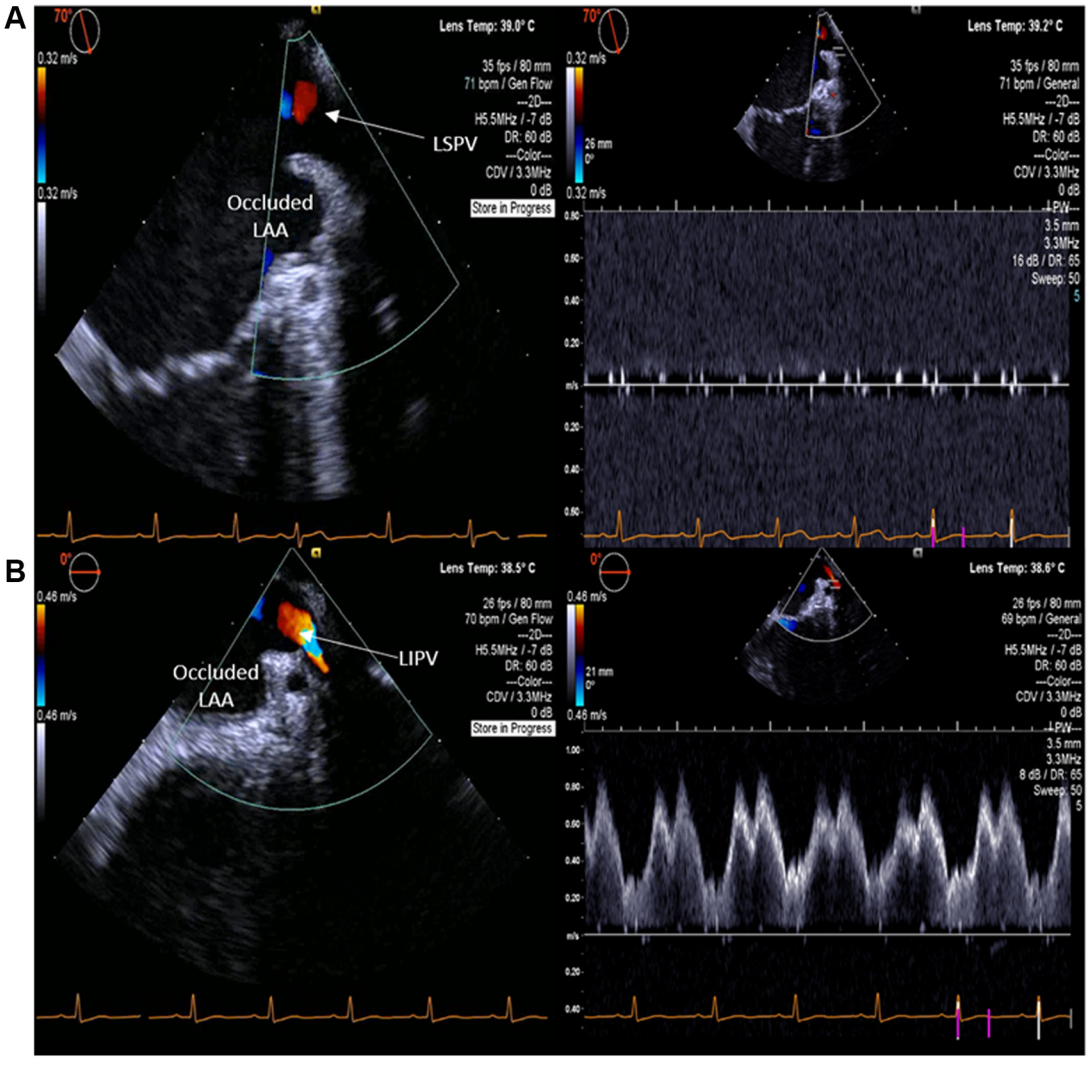
FIGURE 3 Computed Tomography Angiography of the Heart Showing an Occluded Left Superior Pulmonary Vein



(A to D) Coronal section of a computed tomography angiogram showing the left superior pulmonary vein marked by **arrows,** with **(C and D)** occlusion. L = left; R = right.

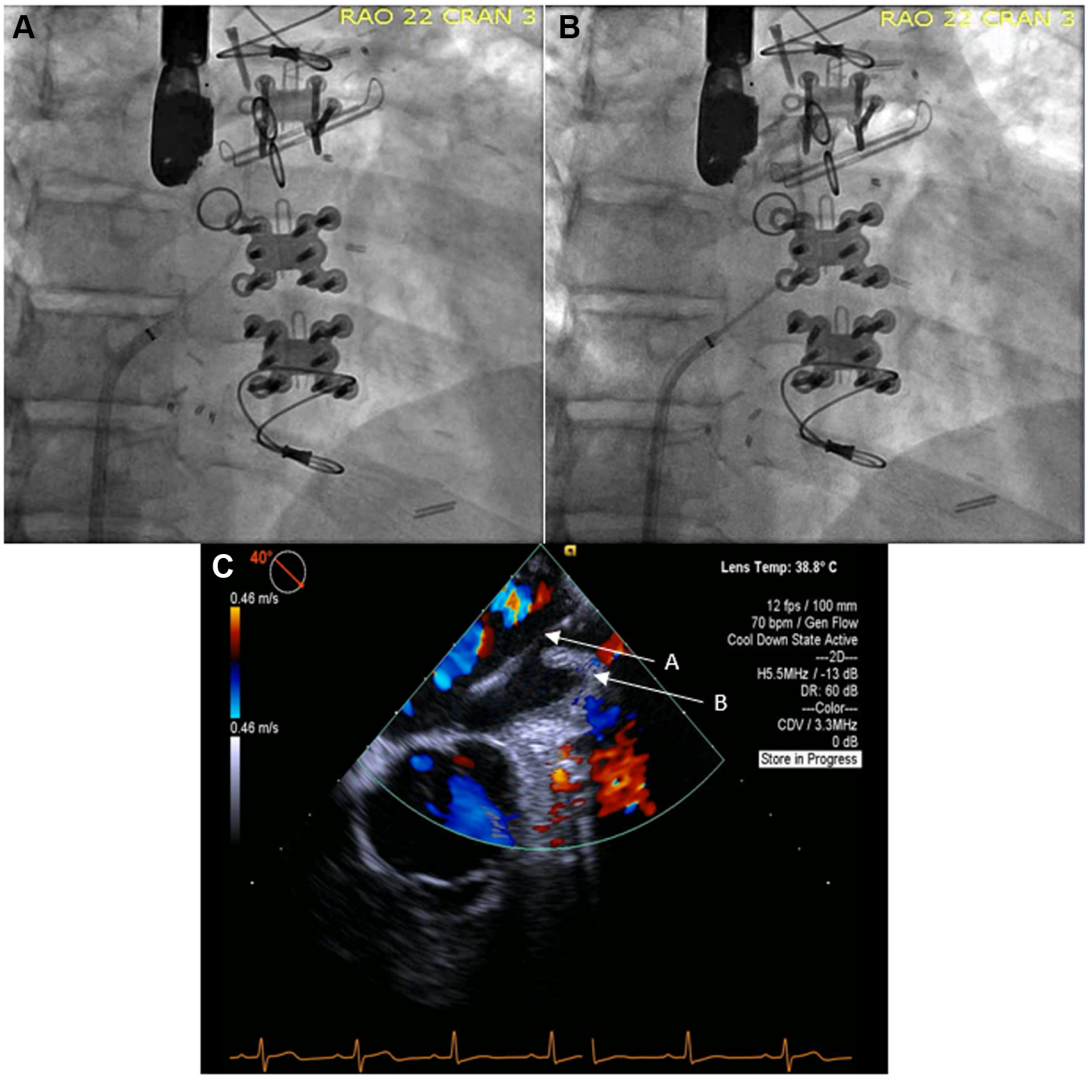
JACC: CASE REPORTS, VOL. 4, NO. 18, 2022
SEPTEMBER 21, 2022:1206–1212

FIGURE 4 Transesophageal Echocardiogram With Pulsed-Wave Doppler Interrogation



Transesophageal echocardiogram and pulsed wave Doppler interrogation showing separation of an occluded left atrial appendage (LAA) and **(A)** the left superior pulmonary vein (LSPV) with no flow and **(B)** the left inferior pulmonary vein (LIPV) with good flow.

FIGURE 5 Precontrast and Postcontrast Angiogram and Intraprocedural Transesophageal Echocardiogram



(A and B) Precontrast and postcontrast angiogram and **(C)** intraprocedural transesophageal echocardiogram showing separation of the left superior pulmonary vein and the left atrial appendage; **A**, catheter in the left superior pulmonary vein; **B**, occluded left atrial appendage. CRAN = cranial; RAO = right anterior oblique.

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 Madhu Reddy, Department of Cardiovascular Medicine, The University of Kansas Health System, 4000 Cambridge Street, Kansas City, Kansas 66160, USA. E-mail: yreddy@kumc.edu. Twitter: @ReddyEPS.

REFERENCES

1. Voeller RK, Zierer A, Schuessler RB, Damiano RJ Jr. Performance of a novel dual-electrode bipolar radiofrequency ablation device: a chronic porcine study. *Innovations (Phila)*. 2011;6(1):17–22.

2. Rostamian A, Narayan SM, Thomson L, Fishbein M, Siegel RJ. The incidence, diagnosis, and management of pulmonary vein stenosis as a complication of atrial fibrillation ablation. *J Interv Card Electrophysiol*. 2014;40(1):63–74.

3. Calkins H, Hindricks G, Cappato R, et al. 2017 HRS/EHRA/ECAS/APHRS/SOLAECE expert consensus statement on catheter and surgical ablation of atrial fibrillation: executive summary. *Europace*. 2018;20(1):157–208.

4. Schoene K, Arya A, Jahnke C, et al. Acquired pulmonary vein stenosis after radiofrequency ablation for atrial fibrillation: single-center experience in catheter interventional treatment. *J Am Coll Cardiol Intv*. 2018;11(16):1626–1632.

5. Pazos-López P, García-Rodríguez C, Guitián-González A, et al. Pulmonary vein stenosis: etiology, diagnosis and management. *World J Cardiol*. 2016;8(1):81–88.

6. Buiatti A, von Olshausen G, Martens E, et al. Balloon angioplasty versus stenting for pulmonary vein stenosis after pulmonary vein isolation for atrial fibrillation: a meta-analysis. *Int J Cardiol*. 2018;254:146–150.

7. Nakaizumi T, Nakamura K, Nakamura K, et al. Haemoptysis and bronchial congestion due to pulmonary vein stenosis after maze procedure. *Respirol Case Rep*. 2019;7(7):e00467.

8. Ayati M, Ouyang F, Kuck K. Pulmonary vein compression after implantation of a left atrial appendage occluder: presentation and discussion of a case. *Indian Pacing Electrophysiol J*. 2014;14(4):194–198.

9. Toale C, Fitzmaurice GJ, Eaton D, Lyne J, Redmond KC. Outcomes of left atrial appendage occlusion using the AtriClip device: a systematic review. *Interact Cardiovasc Thorac Surg*. 2019;29(5):655–662.

KEY WORDS AtriClip, epicardial ablation, Isolator Synergy Ablation System, pulmonary vein stenosis

APPENDIX For a supplemental video, please see the online version of this article.