Acquired Pulmonary Vein Stenosis: One Problem, Two Mechanisms

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Until the last decade, acquired pulmonary vein (PV) stenosis in the adult population was a rare finding, caused by neoplasm or inflammatory conditions such as sarcoidosis or fibrosing mediastinitis. With the increased use of catheter-based ablation for the treatment of atrial fibrillation, PV stenosis is increasingly recognized as a complication of this procedure. Additionally, PV stenosis has been described as a rare complication of cardiac surgery. This report describes two cases of PV stenosis, one acquired as a result of multiple left atrial ablation procedures and the other after surgical cannulation of the right upper PV. (J Am Soc Echocardiogr 2010;23:904.e1-904.e3.)

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Until the last decade, acquired pulmonary vein (PV) stenosis in the adult population was a rare finding, caused by neoplasm or inflammatory conditions such as sarcoidosis or fibrosing mediastinitis. However, with the increased use of catheter-based ablation for the treatment of atrial fibrillation, PV stenosis is increasingly recognized as a complication of this procedure. Additionally, PV stenosis has been described as a rare complication of cardiac surgery. Two cases of acquired PV stenosis are presented.

CASE PRESENTATIONS

Case 1
A 49 year-old man presented with a 13-year history of atrial fibrillation. Other medical history included type 2 diabetes mellitus, hypertension, obstructive sleep apnea, hyperlipidemia, and chronic back pain. After medical therapy for atrial arrhythmias was unsuccessful, the patient underwent 5 radiofrequency ablation procedures over the course of 5 years; the first 4 procedures were for atrial fibrillation, and the most recent was for left atrial flutter. Prior to planned electrical cardioversion for atrial flutter, transesophageal echocardiography (TEE) was performed to exclude left atrial thrombus. Although not well seen on 2-dimensional imaging of the ostium of the left upper PV (Figure 1, Video 1), there was associated color variegating suggestive of flow acceleration on color-flow Doppler imaging (Video 2). Continuous-wave Doppler evaluation revealed a high antegrade flow velocity (maximum, 1.9 m/s) consistent with left upper PV stenosis (mean gradient, 4.8 mm Hg) (Figure 2). Retrospective review of a transthoracic echocardiogram obtained 2 weeks prior to TEE demonstrated turbulent flow in the left atrium in the apical 4-chamber view.

Case 2
A 62-year-old woman underwent TEE after recent aortic valve replacement. Surgery was performed for aortic regurgitation, with the implantation of a 19-mm stentless bioprosthesis. There was no history of transcatheter or surgical therapy for atrial arrhythmias. Her postoperative course was complicated by renal failure requiring hemodialysis and subsequent hospitalization with diarrhea, abdominal pain, shortness of breath, and weakness. She was found to have elevated B-type natriuretic peptide, and echocardiography was performed to assess left ventricular (LV) systolic function. Transthoracic echocardiography revealed normal LV systolic function, systolic narrowing of the LV outflow tract (diameter, 15 mm), and elevated mean and peak LV outflow gradients (23 and 48 mm Hg, respectively). Because the aortic valve prosthesis was not well seen, TEE was performed to evaluate the LV outflow tract and the aortic valve prosthesis.

On TEE, the LV outflow tract was narrow but unobstructed, and the aortic valve prosthesis functioned normally. However, note was made of anatomic narrowing of the ostium of the right upper PV (Figure 3, Video 3) and turbulent antegrade flow entering the left atrium from the right upper PV (Video 4). Spectral Doppler interrogation of PV flow revealed an antegrade peak velocity > 2 m/s. The operative report from the aortic valve replacement procedure was reviewed, confirming placement of a venting cannula in the right upper PV, with no other reference to direct injury to a PV or the left atrium.

DISCUSSION

During the past decade, advances in electrophysiology have led to better understanding of the pathophysiology of atrial fibrillation, and electroanatomic mapping techniques have been developed that allow for catheter-based radiofrequency ablation and electrical isolation of the PV ostia for the treatment of atrial fibrillation and flutter. It is estimated that nearly 50,000 atrial fibrillation ablation procedures are now performed annually in the United States, with clinical success rates reported to be >80%. However, PV stenosis remains a potential complication of catheter ablation for atrial fibrillation, with a current reported incidence of 1% to 3%.1,2
In 2003, Saad et al\textsuperscript{5} reported on 18 patients with severe PV stenosis following catheter ablation of atrial fibrillation. In that study, the median number of stenosed PVs was 2, and total occlusion of a PV was seen in 7 patients. As seen in our first case, stenosis more commonly involved a left-sided PV. The proposed explanation for this was that the ablation catheter can move more easily into a left-sided PV with respiration, and stenosis is more likely when energy is delivered within the PV rather than in the preferred ostial location.

Multidetector computed tomography is considered the gold standard for PV evaluation before and after ablation.\textsuperscript{6} However, even though imaging of the inferior PVs using TEE can be difficult, TEE has been shown to be of value in diagnosing and following patients with PV stenosis. De Piccoli et al\textsuperscript{7} published a series of 96 patients undergoing atrial fibrillation ablation and found that during 1 year of follow-up, PV diameter on TEE decreased by 5% and peak flow velocities increased by 32% in all patients. In those with PV stenoses \( \geq 50\% \) on computed tomography, flow velocities were shown to double over baseline values, and the diameter of the PV on echocardiographic imaging was significantly reduced. Others have shown that severe PV stenosis, as defined at cardiac catheterization, correlates well with a peak velocity \( \geq 2 \) m/s on spectral Doppler interrogation of the PVs.\textsuperscript{8} Although the absolute reduction in PV diameter was not well seen on 2-dimensional images (Figure 1, Video 1) in our first case, the stenosis is readily evident on the basis of color-flow and spectral Doppler evaluation. This example underscores the importance of, and incremental diagnostic value afforded by, careful Doppler examination.

PV stenosis also has been described as a rare complication of cardiac surgery. Most commonly, it is described in the pediatric literature after repair of complex congenital conditions in which the atria and PVs are directly manipulated, such as in anomalous pulmonary venous return or atrial switch operations (the Mustard procedure) in D-transposition of the great vessels.\textsuperscript{9,10} In adults who undergo cardiac surgery, PV stenosis has been reported after autotransplantation for myxoma and after inadvertent injury of the left atrium or PV.\textsuperscript{11,12} However, as a component of cardiopulmonary bypass, a left atrial vent is placed. In patients with significant aortic insufficiency, the right upper PV is a preferred site for cannula placement; when the vent is removed, the defect is sutured. In case 2, suture repair of the right upper PV cannulation site is the most plausible explanation for isolated right upper PV stenosis. To our knowledge, this is the first reported case of PV stenosis acquired by this mechanism.

In conclusion, even as experience increases and interventional techniques improve, acquired PV stenosis remains a potential complication of atrial fibrillation ablation; it also is a rare complication of cardiac surgery. Symptoms can develop even in instances when stenosis is not severe, and lesions can progress over time. Echocardiography has an important role in the diagnosis and follow-up of patients with acquired PV stenosis. Of note, both cases presented here were noted as incidental findings on TEE performed for another indication. Close attention should be paid to pulmonary symptoms in patients with recent ablation or open surgical procedures to facilitate the diagnosis and management of this condition.

REFERENCES


