



Severe hemoptysis as a debut form of left pulmonary vein stenosis

Hemoptisis grave como forma de debut de una estenosis de venas pulmonares izquierdas

Cristina Matesanz López,* Eva Aguilar Rivilla,‡ Diego Monzón Díaz,§
Carmen María Acosta Gutiérrez,¶ María Teresa Río Ramírez¶

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ABSTRACT

Atrial fibrillation is the most common arrhythmia. This pathology carries high healthcare cost and impairs the quality of life of the patients. Percutaneous pulmonary vein ablation is a consolidated therapy to treat it and several studies have shown that long-term recurrence and mortality is lower compared to those who are treated with antiarrhythmic medication. We present the case of a 48-year-old non-smoker man with a clinical history of a difficult-to-control incessant atrial tachycardia that presented to the emergency department with hemoptysis. The clinical picture was due to a complete occlusion of the distal segment of the left pulmonary veins as a complication of a previous percutaneous pulmonary vein ablation. Pulmonary vein stenosis is a rare complication of ablation for atrial fibrillation, being an early diagnosis essential to improve prognosis.

RESUMEN

La fibrilación auricular es la arritmia más común. Es una patología que conlleva un alto coste sanitario y perjudica la calidad de vida de los pacientes. La ablación percutánea de las venas pulmonares es una terapia consolidada para su tratamiento y varios estudios han demostrado que la recidiva a largo plazo y la mortalidad es menor en comparación con pacientes tratados con medicación antiarrítmica. Presentamos el caso de un varón de 48 años, no fumador, con antecedente de taquicardia auricular incesante de difícil control que acude a urgencias por hemoptisis. El cuadro clínico se debió a una oclusión completa del segmento distal de las venas pulmonares izquierdas como complicación de una ablación percutánea previa. La estenosis de la vena pulmonar es una complicación poco común de la ablación para tratar la fibrilación auricular, siendo un diagnóstico precoz fundamental para mejorar el pronóstico.

INTRODUCTION

Atrial fibrillation (AF) is the most common supraventricular tachyarrhythmia. More than 90% of the underlying ectopic foci of electrical activity originate in the pulmonary veins.¹

Percutaneous ablation of the left pulmonary vein (LPV) is a therapeutic alternative in the management of symptomatic recurrent AF and several studies have shown that long-term recurrence and mortality is lower compared to those who are treated with antiarrhythmic medication.² It's a first-line treatment of

patients with symptomatic paroxysmal AF refractory to antiarrhythmic drugs, and offers improved quality of life, although in the case of long-standing persistent AF, percutaneous ablation is more complex and laborious and usually requires more than one intervention.

It is a safe technique but, like any procedure, even in experienced hands it is not exempt from complications, with a rate between 0.8 and 16.3%.³ Complications can be severe, such as cardiac perforation or stroke, or milder, such as those related to the catheter access point.

* Department of Respiratory Medicine. Hospital Universitario de Getafe.

‡ Department of Radiology. Hospital Universitario de Getafe.

§ Department of Cardiac Surgery. Hospital Universitario Gregorio Marañón.

¶ Department of Respiratory Medicine. Hospital Universitario de Getafe.

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CASE PRESENTATION

We present the case of a 48-year-old non-smoker man, chronically anticoagulated due to a difficult-to-control incessant atrial tachycardia. He had required 5 ablation procedures performed in a different hospital: 4 radiofrequency ablations (RFA) and a surgical one. The first four were RFA, which were not effective. The patient was highly symptomatic, describing shortness of breath, anxiety and palpitations due to the AF, reason why he finally undertook a minimal invasive surgery. During the surgery, an Atriclip exclusion device was inserted. He also had suffered from a bradycardia-tachycardia syndrome and was a DDDR pacemaker carrier.

He presented to the emergency department with a 15-day history of hemoptysis and flu-like symptoms the previous month. Sixty milliliters were measured during the first 24 hours at the hospital. On admission, the physical examination was normal and no analytical alterations were observed. The chest X-ray was unremarkable.

During the first hours of hospitalization, hemoptysis increased and the expectoration was more than 150 mL/hour. An emergency bronchoscopy was performed, showing edematous and friable left bronchial tree mucosa with very intense bleeding with the touch of the bronchoscope (*Figure 1*). Instillation of adrenaline was required. The microbiological and pathological analyses of the samples were negative. A chest computed tomography angiography (CT-angiography) was performed, showing both left pulmonary veins (LPV) originated from a common ostium, an occlusion of it and left interstitial edema (*Figure 2*). Our patient had a relevant medical history of cardiac procedures, which was a risk factor to develop this kind of complications.

Given the potential severity of the clinical picture, the patient was referred to another hospital with Cardiac Surgery Department. Once there, a hybrid approach was performed. During the surgery, under extracorporeal circulation, normal drainage of the right pulmonary veins was confirmed in the left atrium. However, the common ostium was completely blocked. Retraction and fibrosis after radiofrequency ablation seemed the etiopathogenic

mechanism, without any relation to the appendage exclusion surgical device. Angioplasty was carried out and weeks later, a stent in the common ostium was placed in a percutaneous procedure with optimal results.

DISCUSSION

The pulmonary veins carry oxygenated blood from the lungs to the heart. The anatomy of the pulmonary veins is variable among patients, with several noteworthy variant and anomalous patterns, including supernumerary pulmonary veins, a common ostium, anomalous pulmonary venous return, and levoatriocardinal veins.⁴ Under normal conditions, four pulmonary veins carry oxygenated blood from both lungs and drain into the left atrium. The right superior pulmonary vein drains the upper and middle lobes, the left superior pulmonary vein drains the upper lobe and lingula, and the two inferior pulmonary veins drain the lower lobes.⁵

The two most common PV anomalies are the presence of a right middle PV and common left trunk.⁶ Our patient not only had a common ostium, but also developed a complete obstruction of it due to previous procedures.

Radiofrequency ablation is an efficacious alternative in patients with symptomatic atrial fibrillation who do not respond to or are intolerant to at least one class I or class III antiarrhythmic drug. Although it is a safe technique,



Figure 1: An edematous and inflamed mucosa was observed in the left upper lobe. It tended to bleed with the touch of the bronchoscope.

pulmonary veins stenosis are still described and it usually occurs about three months after ablation. The severity of PPV stenosis is classified according to the diameter reduction. It is considered slight if the reduction is $< 50\%$, moderate between 50 and 70%, and severe if it is $\geq 70\%$, requiring this last urgent intervention.⁷ Luckily, the incidence has decreased over the last years to between 0.32 and 3.4%.⁸

Patients are usually asymptomatic until they have severe stenosis, as the case we describe.

They often debut in the form of dyspnea or cough from unilateral pulmonary edema or even chest pain. However, hemoptysis is infrequent, as in the previously cited study published by Fender EA et al, where it was only described in 27% of patients with severe stenosis, where pulmonary infarctions can also be observed.⁹

The management is different, depending on the grade of the stenosis. When a 50-70% stenosis is seen, follow-up in 3-6 months is recommended. If it is above 75%, another CT in three months is recommended, unless the stenosis is $> 90\%$, when urgent treatment is required.¹⁰

Our patient presented with severe hemoptysis, which also needs an urgent approach. Hemoptysis develops due to pulmonary venous hypertension. As the resistance of the venous drainage to the left atrium increases, a pulmonary congestion is triggered, moving the plasma fluid from the pulmonary capillaries towards the interstitial spaces and alveoli. That generates pulmonary edema.¹¹

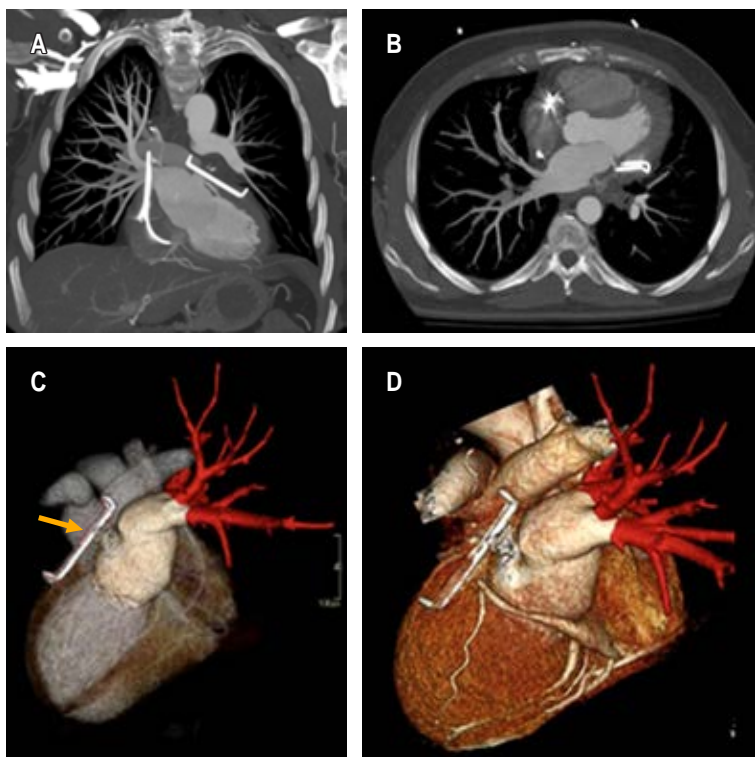


Figure 2: Images corresponding to a computed tomography angiography with intravenous contrast in systemic arterial phase. The greatest contrast opacification can be seen in the pulmonary venous tree and in the systemic arteries. **A)** Axial MPR image with a thickness of 40mm showing the asymmetry of the pulmonary vascularization, as well as the absence of left pulmonary veins draining into the left atrium. **B)** MPR with a thickness of 40 mm showing the same findings as in **A)**. Metal clip on the left appendage. **C)** 3D image of the cardiac volume. Posterior view of the heart. Transparency of cardiac chambers. Left atrium colored white. Right pulmonary veins colored red. Lack of visualization of left pulmonary veins due to its obstruction and therefore absence of contrast within it. Clip on left appendage indicated by arrow. Ascending aorta and pulmonary arteries in gray. **D)** 3D image similar to the previous one. Volumetric reconstruction of the cardiac surface. Posterior view of the heart. MPR = multiplanar reconstruction.

CONCLUSIONS

Pulmonary vein stenosis related to radiofrequency ablation is becoming a less frequent complication, which makes it sometimes hard to diagnose. Our patient had a medical history of AF ablations and surgical cardiac procedures. This should be always taken into account in order to be able to suspect this pathology soon and avoid unnecessary delay in the diagnosis.

Although it is a well described complication, it is rare and physicians that do not usually work in that area are frequently unaware of it. Moreover, since symptoms are not specific, they could be mistaken.

Hemoptysis, therefore, may be the first manifestation of a large number of diseases, and not only lung ones. Medical history may be the key to focus the diagnosis.

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Correspondence:

Cristina Matesanz López

Department of Respiratory Medicine. Hospital Universitario de Getafe

E-mail: cristina.matesanz@salud.madrid.org