

Partially reversible lung consolidation after revascularization of a total occlusion of both left pulmonary veins following ablation of atrial fibrillation: a case report

Anete Ahero ^{1*}, Thomas Frauenfelder², Alexander Breitenstein ³, Peter Ammann⁴, Nils Kucher⁵, and Stefano Barco ⁵

¹Clinic for Internal Medicine, University Hospital Zurich, Raemistrasse 100, 8091 Zürich, Switzerland; ²Institute for Diagnostic and Interventional Radiology, Raemistrasse 100, 8091 Zürich, Switzerland; ³Clinic for Cardiology, University Hospital Zurich, Raemistrasse 100, 8091 Zürich, Switzerland; ⁴Clinic for Cardiology, Cantonal Hospital St. Gallen, Rorschacher Str. 95/Haus 01, 9007 St. Gallen, Switzerland; and ⁵Clinic for Angiology, University Hospital Zurich, Raemistrasse 100, 8091 Zürich, Switzerland

Received 9 December 2021; first decision 31 December 2022; accepted 31 January 2023; online publish-ahead-of-print 2 February 2023

Background

The use of pulmonary vein (PV) radiofrequency ablation for atrial fibrillation (AF) treatment may be complicated by PV stenosis or occlusion. A common curative treatment for symptomatic patients is a transcatheter intervention, including percutaneous transluminal balloon angioplasty and stent implantation. Stent implantation itself, however, can be complicated by in-stent stenosis.

Case summary

A 26-year-old man presented with worsening exertional dyspnoea due to a total occlusion of both left PVs after the isolation of two PVs for AF. Chest computed tomography (CT) showed chest asymmetry and consolidation of the left lung. The patient was treated with balloon angioplasty and stent placement of both left PVs, resulting in improvement of symptoms, walking distance, and increase in lung space volume by 120 mL based on CT-based volumetry. Ten months later, the patient experienced a recurrence of similar symptoms. A high grade in stent restenosis of the upper left PV and moderate in stent restenosis of the lower PV were diagnosed and treated with angioplasty. The patient was discharged from the hospital in good clinical condition 3 days after the intervention.

Discussion

Non-specific symptoms of PV stenosis or occlusion, such as shortness of breath, fatigue, flu-like symptoms, reduced physical performance, and haemoptysis delay the diagnosis. If unusual symptoms appear abruptly after PV isolation, a PV stenosis should be considered. In this case, we describe for the first time a partially reversible consolidation of lung parenchyma following the revascularization of both PVs.

Keywords

Pulmonary vein ablation • Pulmonary vein occlusion • Pulmonary vein angioplasty • Lung volume consolidation • Case report

ESC Curriculum 5.3 Atrial fibrillation • 6.5 Cardiomyopathy • 8.6 Secondary prevention

Learning points

- A total occlusion of both left pulmonary veins (PVs) may not necessarily lead to pulmonary infarction, most likely due to the development of collateral bronchial and epigastric draining veins.
- Lung space volume may increase after revascularization of the PVs. The correlation of lung space volume and revascularization has to be further examined.

* Corresponding author. Tel: +41 78 403 38 59, Email: anete.ahero@gmail.com

Handling Editor: Rami Riziq Yousef Abumualeq

Peer-reviewers: George K. Andrikopoulos; Henning Jansen; Fabian Barbieri

Compliance Editor: Zhiyu Liu

Supplementary Material Editor: Niklas Shenker

© The Author(s) 2023. Published by Oxford University Press on behalf of the European Society of Cardiology.

This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial License (<https://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

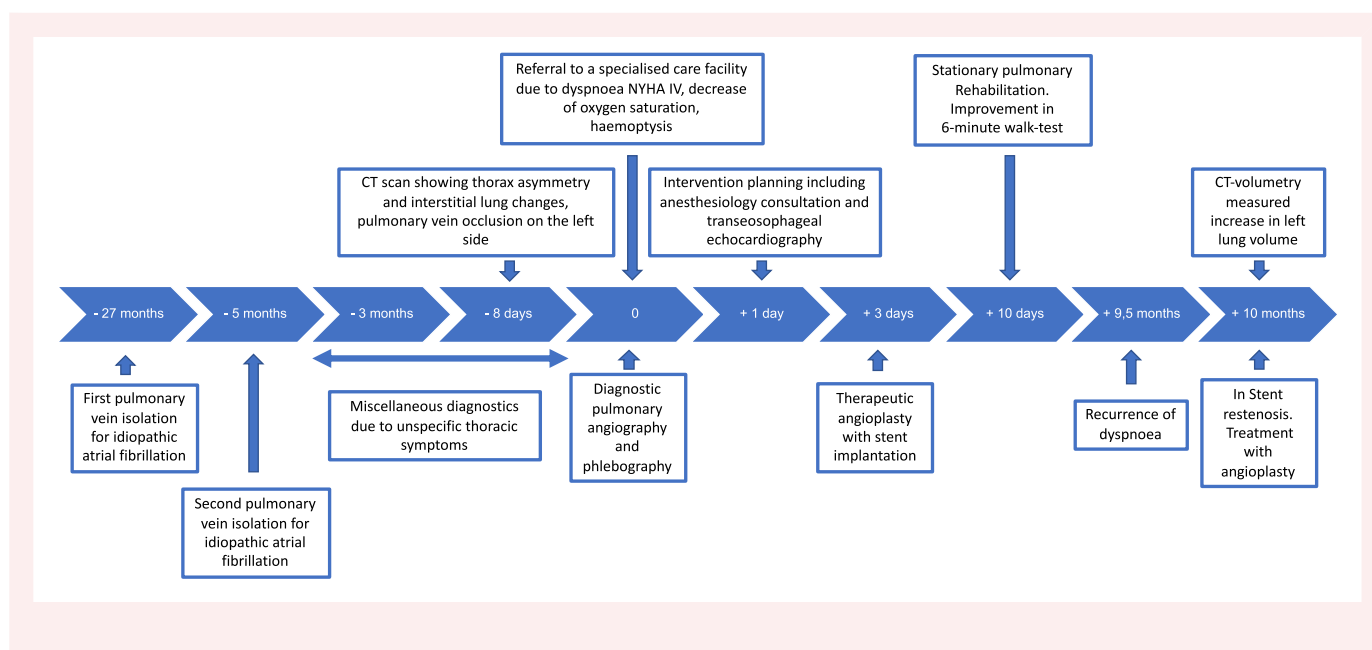
Primary specialities involved other than cardiology

Angiology, radiology, internal medicine.

Introduction

The use of pulmonary vein (PV) radiofrequency ablation (RFA) for atrial fibrillation (AF) treatment is complicated by PV stenosis (PVS) or occlusion (PVO) in 0.3–3.4% of cases according to the available literature.^{1–5} Typical symptoms are shortness of breath, fatigue, flu-like symptoms, reduced physical performance, and haemoptysis. Furthermore, recurrent pneumonia, pleural effusion, pulmonic parenchymal consolidation, and pulmonic hypertension may result in severe functional impairment.^{1–11} Since most patients are referred to a general practitioner or pneumologist for these symptoms, it is of paramount interest for the patient and the physicians to be aware of that. A common curative treatment for symptomatic patients is represented by transcatheter interventions, including percutaneous transluminal balloon angioplasty (PTA) and stent implantation.^{2,4–6,8} The risk of PV restenosis may depend on the type of endovascular treatment that has been performed. There is evidence indicating that stent implantation with balloon dilatation is characterized by a better short-term outcome and lower risk of PV restenosis compared with PTA alone.^{2,6} Indeed, more than 50% of patients will experience an early restenosis after PTA alone,^{2,4,6} whereas this early risk is much lower with 16–33% after a stent placement depending on the stent size. Large-diameter stents (≥ 8 mm) may have a lower risk of restenosis (6.4%).^{2,6} The decision related to continuation or beginning of anticoagulation in addition to antiplatelet agents after the PV stent implantation depends on congestive heart failure, hypertension, age, diabetes mellitus, prior stroke or transient ischaemic attack or thromboembolism, vascular disease, age, sex category (CHA₂DS-VAS_c score, as there are no specific guidelines regarding this.

Timeline



Case

A 26-year-old patient with lone AF and a history of tachycardia induced cardiomyopathy was referred to a tertiary care facility due to worsening dyspnoea and haemoptysis after two PV isolations (PVI) for idiopathic AF (first PVI 27, second PVI 5 months before referral). The examinations that were previously conducted at another institution included computed tomography (CT) scan, chest ultrasound, pleural puncture, bronchoscopy, and positron emission tomography CT scan. An obvious reason explaining the symptoms could not have been revealed as the patient came to our attention for a second assessment. The patient presented with dyspnoea New York Heart Association functional classification (NYHA) IV and intermittent decrease of oxygen saturation (nadir 89%). Auscultatory normal breathing sounds seemed present, without any further abnormal findings in the physical examination. Laboratory findings demonstrated an insignificant increase of N-terminal pro-b-type natriuretic peptide (NT-proBNP) at 99 ng/L (normal range <85.8 ng/L). The patient was otherwise healthy with no known cardiovascular risk factors. A new episode of AF has not been documented since the second PVI.

The patient was diagnosed with PVO of both left-sided PVs. A CT scan showed thorax asymmetry and interstitial lung changes with ground-glass opacities as well as a complete occlusion of the left pulmonary veins (Figure 1A). Transthoracic echocardiography and cardiac magnetic resonance imaging (MRI) demonstrated a slightly restricted left ventricular ejection fraction (48%) with otherwise non-pathological cardiac MRI. The pulmonary angiogram showed slow flow in the left pulmonary artery; no venous backflow was detectable in the PVs. However, a backflow was documented in bronchial as well as epigastric veins. Invasive pulmonary artery pressure was 40/22 mmHg (mean 31 mmHg) on the left and 34/22 mmHg (mean 28 mmHg) on the right side, compatible with moderate pulmonary hypertension [World Health Organization (WHO) Class I—due to veno-occlusive disease].

The occluded left lower PV was treated with a drug eluting stent 5.0 × 30 mm and an 8 × 27 mm bare metal stent served to treat the PV obstruction in the upper PV using a transeptal approach. Due to CHA₂DS-VAS_c score of 0 points, a dual antiplatelet therapy with aspirin 100 mg orally once a day and clopidogrel 75 mg orally once a day was

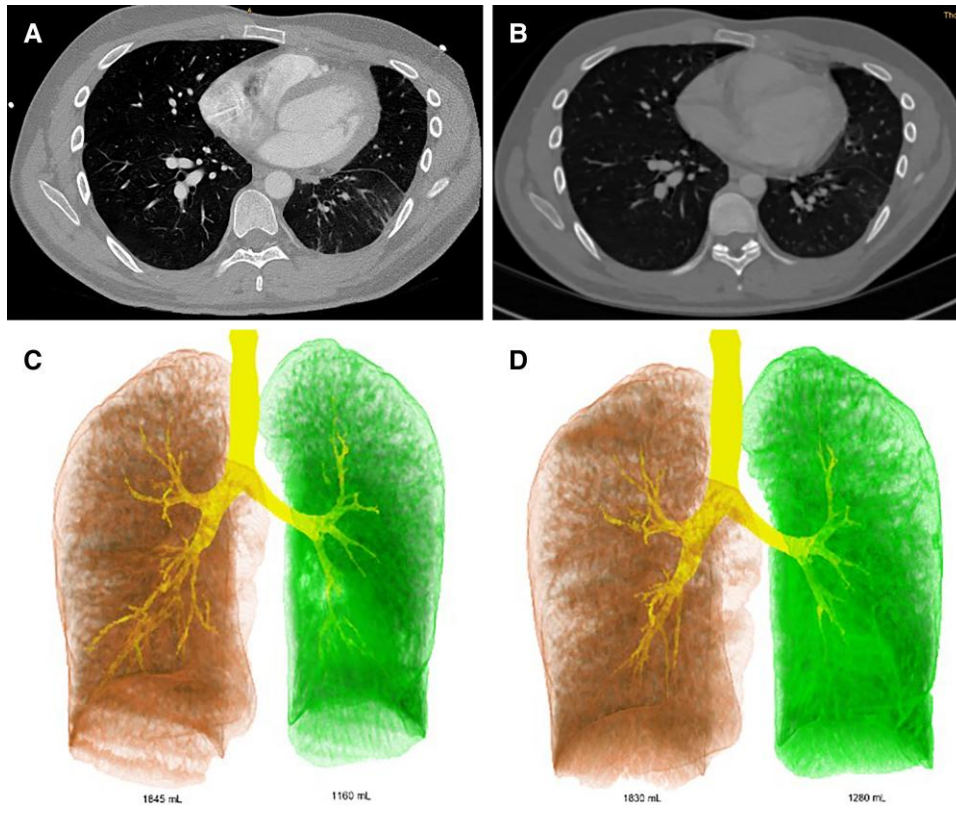


Figure 1 Lung space volumes pre and post-revascularization of both pulmonary veins. (A) Axial contrast enhanced computed tomography pre-intervention showing left-sided lung consolidation and chest asymmetry. (B) An improvement of lung volume and chest asymmetry after intervention. (C) Lung volumetry by chest computed tomography pre-intervention. (D) Lung volumetry by chest computed tomography post-intervention showed an increase in lung volume by 120 mL.

administered after stent implantation for 3 months followed by aspirin 100 mg once a day alone. The patient did not receive any other medications. The patient showed immediate clinical improvement of dyspnoea. Furthermore, after rehabilitation, a 6 min walk-test confirmed an improvement in walking distance from 25 to 430 m without desaturation of the peripheral blood oxygenation. Transthoracic echocardiography confirmed normalized pulmonary pressure. Furthermore, the left lung volume increased from 1160 to 1280 mL based on CT volumetry. Total lung space volume was 3005 mL before and 3110 mL after intervention (Figure 1). Based on these measurements, an increase in functioning lung tissue volume could be detected.

Ten months later, the patient experienced a recurrence of similar symptoms. A high grade in stent restenosis of the upper left PV and moderate in stent restenosis of the lower PV were diagnosed and treated with angioplasty. The patient was discharged from the hospital in good clinical condition 3 days after the intervention.

Discussion

Atrial fibrillation represents the most common arrhythmic disease. The predicted incidence is 17.9 million people in Europe by 2060 and 6–12 million people in the USA by 2050, with prevalence increasing from 0.5% at 50–59 years to 10% at ≥ 80 years.^{12,13} Radiofrequency ablation is one of the therapeutic options to treat AF, with a success rate of

~75%.^{14–16} Pulmonary vein stenosis as a complication of the RFA occurs due to thermal injury in the tissue surrounding the area. Risk of PV stenosis has been reduced over the years from >6 to $<1\%$.^{1–5} The risk of re-stenosis by stent implantation varies from 16 to 33% with a significantly lower risk if a stent with diameter ≥ 8 mm is being implanted after PTA only, the risk of restenosis varies between 53 and 83%.^{2,4,6} New therapeutic approaches are being developed. These may minimize the risk of complications. As an example, a pulsed field ablation has shown to potentially reduce the risk of PVS (with diameter reduction of only 10–20% of a vessel) as well as of other complications associated with RFA in a canine model¹⁷ and should be examined further.

Due to a usually mild presentation of the symptoms, the average time until diagnosis ranges from 3 to 7 months.^{7,10,11} Major factors for the delay in diagnosis are non-specific symptoms such as shortness of breath, fatigue, flu-like symptoms, reduced physical performance, and haemoptysis. Recurrent pneumonia, pleural effusion, pulmonary parenchymal consolidation, and pulmonary hypertension may occur. In patients who have undergone PVI and present with exertional dyspnoea or haemoptysis, PVS should be considered.^{1–11}

In this case, there was a delay in the diagnosis of PVS of 3 months. After the procedure, the patient experienced improvement of exertional dyspnoea from NYHA IV to NYHA II immediately, it further improved to NYHA I after rehabilitation. We describe for the first time partially reversible consolidation of lung parenchyma by CT-based

volumetry following the revascularization of both PVs. These findings suggest that (i) a total occlusion of both left PVs may not necessarily lead to pulmonary infarction, most likely due to the development of collateral bronchial and epigastric draining veins and (ii) that lung space volume may increase after revascularization of the PVs. The correlation of lung space volume and revascularization has to be further examined. Our patient received aspirin after stent implantation. It remains unclear whether the onset of such a complication may be possibly related to a specific antithrombotic therapy or regimen, particularly among patients with no indication to receive oral anticoagulation.

Lead author biography



Anete Ahero is currently a resident in internal medicine. She obtained the medical doctor degree at Riga Stradins University, Latvia, in 2015. She studied in an ERASMUS program for a year at the University of Münster, Germany, in 2013/2014. She started her internal medicine residency in Spital Zollikerberg, Switzerland, 2017–19, which she continued at the University Hospital Zurich, Switzerland.

Supplementary material

Supplementary material is available at *European Heart Journal – Case Reports*.

Acknowledgements

None declared.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The patient signed the Informed Consent Form on 23 March 2021, where it is stated that he received the information sheet, was adequately informed, consented health-related data (including genetic data) and biological material to be used for research purposes. The authors confirm that written consent for submission and publication of this case report including the images and associated text have been obtained from the patient in line with COPE guidance.

Conflict of interest: There are no relevant financial disclosures in relation to the present article. A.B. received consulting fees from Abbott, Bayer Health Care, Biosense Webster, Biotronik, BMS/Pfizer, Boston Scientific, Cook Medical, Daiichi Sankio, and Medtronic. A.B. received

educational grants from Biosense Webster, Biotronik, and Actelion. A.B. received presenter fees from Abbott, Bayer Health Care, Biotronik, Cook Medical, BMS/Pfizer, Boston Scientific, Daiichi Sankio, Medtronic, and Spectranetics/Philippis.

Funding: None declared.

References

- Teunissen C, Velthuis BK, Hassink RJ, van der Heijden JF, Voncken E-JPA, Clappers N, et al. Incidence of pulmonary vein stenosis after radiofrequency catheter ablation of atrial fibrillation. *JACC Clin Electrophysiol* 2017;**3**:589–598.
- Schoene K, Arya A, Jahnke C, Paetsch I, Nedios S, Hilbert S, et al. Acquired pulmonary vein stenosis after radiofrequency ablation for atrial fibrillation single-center experience in catheter interventional treatment. *JACC Cardiovasc Interv* 2018;**11**:1626–1632.
- 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**:63–74.
- Fender EA, Widmer RJ, Hodge DO, Packer DL, Jr HDR. Assessment and management of pulmonary vein occlusion after atrial fibrillation ablation. *JACC Cardiovasc Interv* 2018;**11**:1633–1639.
- Raeisi-Giglou P, Wazni OM, Saliba WI, Barakat A, Tarakji KG, Rickard J, et al. Outcomes and management of patients with severe pulmonary vein stenosis from prior atrial fibrillation ablation. *Cir Arrhythm Electrophysiol* 2018;**11**:e006001.
- Li Y-J, Pan X, Wang C, He B. Stent implantation for severe pulmonary vein stenosis or occlusion secondary to atrial fibrillation ablation. *Int J Cardiol* 2020;**301**:85–89.
- Fender EA, Widmer RJ, Hodge DO, Cooper GM, Monahan KH, Peterson LA, et al. Severe pulmonary vein stenosis resulting from ablation for atrial fibrillation: presentation, management, and clinical out-comes. *Circulation* 2016;**134**:1812–1821.
- Ernst S, Ouyang F, Goya M, Löber F, Schneider C, Hoffmann-Riem M, et al. Total pulmonary vein occlusion as a consequence of catheter ablation for atrial fibrillation mimicking primary lung disease. *J Cardiovasc Electrophysiol* 2003;**14**:366–370.
- Prieto LR, Kawai Y, Worley SE. Total pulmonary vein occlusion complicating pulmonary vein isolation: diagnosis and treatment. *Heart Rhythm* 2010;**7**:1233–1239.
- Packer DL, Keelan P, Munger TM, Breen JF, Asirvatham S, Peterson LA, et al. Clinical presentation, investigation, and management of pulmonary vein stenosis complicating ablation for atrial fibrillation. *Circulation* 2005;**111**:546–554.
- Qureshi AM, Prieto LR, Latson LA, Lane GK, Igor Mesia C, Radvansky P, et al. Transcatheter angioplasty for acquired pulmonary vein stenosis after radiofrequency ablation. *Circulation* 2003;**108**:1336–1342.
- Yiin GSC, Howard DPJ, Paul NLM, Li L, Luengo-Fernandez R, Bull LM, et al. Age-specific incidence, outcome, cost, and projected future burden of atrial fibrillation-related embolic vascular events: a population-based study. *Circulation* 2014;**130**:1236–1244.
- Morillo CA, Banerjee A, Perel P, Wood D, Jouven X. Atrial fibrillation: the current epidemic. *J Geriatr Cardiol* 2017;**14**:195–203.
- Cappato R, Calkins H, Chen S-A, Davies W, Lesaka Y, Kalman J, et al. Worldwide survey on the methods, efficacy and safety of catheter ablation for human atrial fibrillation. *Circulation* 2005;**111**:1100–1105.
- Fisher JD, Spinelli MA, Mookherjee D, Krumerman AK, Palma EC. Atrial fibrillation ablation: reaching the mainstream. *Pacing Clin Electrophysiol* 2006;**29**:523–537.
- Callans DJ, Gerstenfeld EP, Dixit S, Zado E, Vanderhoff M, Ren JF, et al. Efficacy of repeat pulmonary vein isolation procedures in patients with recurrent atrial fibrillation. *J Cardiovasc Electrophysiol* 2004;**15**:1050–1055.
- Howard B, Haines DE, Verma A, Packer D, Kirchner N, Barka N, et al. Reduction in pulmonary vein stenosis and collateral damage with pulsed field ablation compared with radiofrequency ablation in a canine model. *Circ Arrhythm Electrophysiol* 2020;**13**:e008337.