Case Report

Endovascular transatrial stenting of pulmonary vein stenosis after lung transplantation

Michaela Orlitová 1,2, Marc Gewillig 2,3, Jan Van Slambrouck 1,4, Dirk Vlasselaers 5,6, Bart Jacobs 5,6, Arne P. Neyrinck 2,7, Lieven Depypere 1,4, Laurent Godinas 5,8, Robin Vos 4,8, Geert M. Verleden 4,8, Dirk E. Van Raemdonck 1,4, Laurens J. Ceulemans 1,4,*

1 Department of Thoracic Surgery, University Hospitals Leuven, Leuven, Belgium
2 Department of Cardiovascular Sciences, KU Leuven, Leuven, Belgium
3 Department of Pediatric and Congenital Cardiology, University Hospitals Leuven, Leuven, Belgium
4 Department of Chronic Diseases and Metabolism, Laboratory of Respiratory Diseases and Thoracic Surgery (BREATHE), KU Leuven, Leuven, Belgium
5 Department of Intensive Care Medicine, University Hospitals Leuven, Leuven, Belgium
6 Department of Cellular and Molecular Medicine, KU Leuven, Leuven, Belgium
7 Department of Anesthesiology and Algology, University Hospitals Leuven, Leuven, Belgium
8 Department of Respiratory Diseases, University Hospitals Leuven, Leuven, Belgium

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ABSTRACT

Pulmonary vein stenosis (PVS) and pulmonary vein occlusion (PVO) represent rare complications after lung transplantation (LTx), with limited therapeutic options and a high risk of graft loss. We present 2 cases of successful endovascular transatrial stenting following double LTx. A 60-year-old woman with chronic obstructive pulmonary disease who underwent double lobar LTx was diagnosed at postoperative day 72 with a high-grade PVS on the left side. A 22-year-old woman with idiopathic pulmonary arterial hypertension who underwent double LTx was diagnosed 9 days later with PVO of the left upper lobe vein. To avoid surgical reintervention, endovascular transatrial dilatation and stenting were performed successfully in both cases. Transatrial endovascular stenting of PVS or PVO after LTx seems an effective and safe treatment option that should be considered for these life-threatening complications and executed with care.

1. Introduction

Pulmonary vein (PV) anastomosis in lung transplantation (LTx) can be complicated by pulmonary vein stenosis (PVS) or pulmonary vein occlusion (PVO) due to compression, kinking, or vascular wall injury. PVS and PVO may result in thrombosis with increased capillary congestion and ventilation/perfusion (V/Q) mismatch resulting in hypoxemia, edema, pleural effusion, or hemodynamic collapse.

PVS represents a rare but life-threatening complication after LTx, with a prevalence of 1.4% and mortality of up to 45%.1 Diagnosing PVS remains challenging owing to its clinical presentation resembling primary graft dysfunction (PGD), infection, or acute rejection. Standard diagnostic modalities include transesophageal echocardiography (TEE), computed tomography angiography (CTA), or magnetic resonance imaging. Treatment options include open repair, lobectomy/pneumonectomy, or retransplantation. Although an endovascular approach for PVS following catheter ablation for atrial fibrillation is well-described,2 angioplasty with stenting has rarely been reported after LTx.3-8

We present 2 cases of PVS and PVO following double LTx, successfully treated with endovascular transatrial stenting, and provide an overview of the reported experience after LTx. The ethics committee of University Hospitals Leuven approved reporting (S51577).

Abbreviations: CTA, Computed tomography angiography; LA, Left atrium; LL, Left lung; LTx, Lung transplantation; LULV, Left upper lobe vein; PGD, Primary graft dysfunction; PG, Peak instantaneous gradient; POD, Postoperative day; PV, Pulmonary vein; PVO, Pulmonary vein occlusion; PVS, Pulmonary vein stenosis; TEE, Transesophageal echocardiography; V/Q, Ventilation/perfusion.

* Corresponding author. Herestraat 49, 3000 Leuven, Belgium.
E-mail address: laurens.ceulemans@uzleuven.be (L.J. Ceulemans).
@CeulemansLJ (L.J. Ceulemans)

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2. Case report

2.1. Case 1

A 60-year-old woman (height, 163 cm; weight, 58 kg) with chronic obstructive pulmonary disease underwent double lobar LTx (right lower/ middle lobe and left lower lobe) in July 2020 under venoarterial extracorporeal membrane oxygenation support. The donor was a brain-dead 32-year-old man (height, 190 cm; weight, 95 kg).

PGD at 0, 24, 48, and 72 hours after LTx was 2, 2, 2, and 1, respectively. The patient could not be weaned off the ventilator, requiring tracheostomy on the postoperative day (POD) 11. Frequent left lung (LL) aspiration was performed for impaction. On POD 53, bilateral thoracic drainage was required for pleural effusion, and TEE 5 days later revealed left PVS, confirmed using CTA (residual diameter: 3.7 mm over 6 mm) (Fig. A, B). Right/left perfusion scintigraphy was 88%/12%.

As the patient remained stable, the initial treatment was conservative. However, intermittent mechanical ventilation was continuously needed. Therefore, an endovascular approach was performed on POD 131. An 8-11-F sheath was placed in the right femoral vein. Through the right atrium, a transatrial puncture was performed. The solitary left PV was passed using a Progreat Micro Catheter (Terumo) and 4-F vertebral catheter. Interrogation with 12-mm balloon dilatation (4401204X Powers, Cordis) was performed; subsequently, a 26-mm stent (EV3 Intrastent-LDMAX; S18-26, Medtronic) was deployed up to 12 mm.

An immediate hemodynamic response was observed, and the peak pulmonary velocity over stenosis dropped from 147 cm/s (Fig. C) to 60 cm/s. A pressure Peak instantaneous gradient (PG) between PV and left atrium (LA) measured before stenting was 12 mmHg, which restored to laminar flow without significant pressure PG after stenting. Acetylsalicylic acid at 80 mg was initiated.

Seven days after intervention, the patient was definitively extubated and discharged from the intensive care unit on POD 152 and from the hospital on POD 176. Follow-up computed tomography and V/Q scans obtained 65 days after stenting showed an in-stent diameter of 10 mm (Fig. D) and right/LL perfusion of 59%/41%. At the last follow-up (POD 550), pulmonary function remained stable with a forced vital capacity of 1.86 L/60% and a forced 1-second expiratory volume of 1.67 L/68%.

2.2. Case 2

A 22-year-old woman (height, 163 cm; weight, 74 kg) with pulmonary hypertension underwent double LTx in October 2021 under venoarterial extracorporeal membrane oxygenation support. The donor was a brain-dead 27-year-old woman (height, 159 cm; weight, 63 kg). LL implantation was challenged by a hypertrophic heart, and the PV bifurcation was too proximal for atrial anastomosis, for which both recipient's veins were reunited to create a single cuff (Fig. E).

Due to bilateral pulmonary edema and limited cardiac function, extracorporeal membrane oxygenation was continued for 2 days (PGD3 at all time points). Although the right ventricle recuperated, the patient could not be weaned off ventilation and the LL remained consolidated, accompanied by pleural effusion (Fig. F). CTA on POD 5 revealed an LA thrombus but patent pulmonary veins. A V/Q scan obtained on POD 9 showed 22.5% LL perfusion with upper/lower lobe distribution of 5.6%/16.9%. On POD 10, right-side heart catheterization and transatrial puncture via a 6-11-F sheath in the right femoral vein was performed. Occlusion of the left upper lobe vein (LULV) was diagnosed (Fig. G).

After interrogation, a kink, distal of the anastomosis, was passed with a guidewire (Terumo), revealing a patent PV filled with thrombi (Fig. H). A 12-mm balloon (Tyshack, Braun) interrogation was performed, which revealed safe stretchability up to 10 mm; a 10/20-mm stent (Cook Medical) was implanted.

The peak pulmonary velocity over stenosis dropped from 172 cm/s before stenting to 77 cm/s after stenting. The PG between LULV and LA before stenting was 12 mmHg, which restored to laminar flow. Low-molecular-weight heparin (Enoxaparin 60 mg) was initiated and switched to oral anticoagulants (Edoxaban 30 mg) for deep venous thrombosis.

A tracheostomy was placed on POD 18, and the patient became ventilator independent on POD 36; she was discharged from the intensive care unit 5 days later and from the hospital on POD 61. Follow-up contrast injection, visualization of left upper lobe vein with thrombi after pulmonary vein occlusion passing. CTA, computed tomography angiography; Vmax, maximum velocity; Vmean, mean velocity; PG, peak instantaneous gradient; VTI, velocity time integral.

Fig. Case 1: (A) Coronal plane CTA on the postoperative day 72, bilateral (Left more pronounced) lung consolidation and pleural effusion. (B) Transverse plane CTA, arrow: pulmonary vein stenosis. (C) Transthoracal echocardiography depicting increased pulmonary velocity over stenosis. (D) Transverse plane CTA performed 65 days after intervention, arrow: stent. Case 2: (E) Heart catheterization, visualization of side-to-side recipient venous anastomosis, creating a “donut-like” shape when anastomosed end-to-end with the donor atrial cuff. (F) Coronal plane CTA on the postoperative day 5, left lung consolidation, and pleural effusion concentrated in the left upper lobe. (G) Heart catheterization contrast injection visualized only lower pulmonary vein due to left upper lobe vein occlusion. (H) Heart catheterization contrast injection, visualization of left upper lobe vein with thrombi after pulmonary vein occlusion passing. CTA, computed tomography angiography; Vmax, maximum velocity; Vmean, mean velocity; PG, peak instantaneous gradient; VTI, velocity time integral.
computed tomography and V/Q scans obtained 83 days after stenting confirmed a patent LULV with 10.5-mm diameter and LL (upper/lower lobe distribution) perfusion of 42% (19%/23%), respectively. Pulmonary function at the last follow-up (POD 133) remained stable with a forced vital capacity of 1.72 L/45% and a forced 1-second expiratory volume of 1.46 L/44%.

3. Discussion

We describe 2 cases of successful transatrial endovascular stenting of the left PV after LTx, avoiding surgical reintervention.

Eight other cases of endovascular PVS treatment, all isolated LTx, have been reported (Table).3,8,10,12 Most common clinical presentations were respiratory insufficiency, pulmonary edema, or pleural effusion. In 89% (8/9) of the cases, CTA confirmed PVS at a mean of 215 days after LTx. All reported PV diameters were <4 mm. The mean pressure gradient over PVS was 18 mmHg. The diameter of all implanted stents was 2-10 mm. Stenting was performed in 78% (7/9) of the cases. Antithrombotic strategy ranged from monotherapy to triple therapy. Symptoms resolved in 78% (7/9) of the cases and recurred in 2 patients who died.

These findings suggest that single LTx might have a higher risk of developing symptomatic PVS than double LTx, owing to limited compensation by the contralateral diseased lung. Apart from optimization of caliper mismatch, surgical strategies to avoid PVS/PVO are stay sutures for orientation, prevention of kinking by limiting the vessel length, and techniques for resizing short atrial cuffs.13,14

According to the literature, PVS after LTx could be considered from a peak PV velocity of 100 to 120 cm/s (mean reported velocity, 208 ± 86 cm/s) and a diameter of <5 mm or 50% reduction in comparison to the contralateral PV.11,15 Contributing factors to the development of PVS/PVO could be changing intrathoracic pressures, heart remodulation, edema, infection, or hemorrhagic complications. Likely, the PVO in case 2 occurred with right ventricle normalization after LTx.

Early PVS/PVO is challenging to diagnose because of its clinical presentation resembling PGD, infection, or acute rejection. Late post-transplant PVS can resemble infection or allograft rejection. Early imaging by CTA, TE, magnetic resonance imaging, heart catheterization, or V/Q scan is critical.

Management ranges from conservative to open repair, graft removal, retransplantation, or endovascular stenting. A conservative approach (pharmacoeffective management of edema, intensive rehabilitation, and

<table>
<thead>
<tr>
<th>Table</th>
<th>Literature review.</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PVS</strong></td>
<td></td>
</tr>
<tr>
<td>Clark et al. (1996)</td>
<td>F, 32</td>
</tr>
<tr>
<td>Zimmermann et al. (2009)</td>
<td>M, 42</td>
</tr>
<tr>
<td>Pazos-López et al. (2010)</td>
<td>F, 31</td>
</tr>
<tr>
<td>Loyalka et al. (2012)</td>
<td>M, 56</td>
</tr>
<tr>
<td>Mohamed Mydlin et al. (2012)</td>
<td>M, 63</td>
</tr>
<tr>
<td>Rashad et al. (2015)</td>
<td>F, 64</td>
</tr>
<tr>
<td>Johanputra et al. (2017)</td>
<td>F, 60</td>
</tr>
<tr>
<td>Jing et al. (2021)</td>
<td>M, 31</td>
</tr>
<tr>
<td><strong>PVO</strong></td>
<td></td>
</tr>
<tr>
<td>Orlitová et al., case 1</td>
<td>F, 60</td>
</tr>
<tr>
<td>Orlitová et al., case 2</td>
<td>F, 22</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Reference</th>
<th>PPCCFV (mmHg)</th>
<th>PV PG (mmHg)</th>
<th>Time of stenting</th>
<th>Type of stent (mm)</th>
<th>Antithrombotic therapy</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>PVS</strong></td>
<td></td>
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<tr>
<td>Clark et al. (1996)</td>
<td>Information</td>
<td>Information</td>
<td>Several mo post-LTx</td>
<td>Wall stent</td>
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<td>21 mo after LTx: RF, GIT bleed</td>
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<td>Information</td>
<td>Information</td>
<td>16 d after LTx</td>
<td>⊗ 10 × 16</td>
<td>100 mg/d A + 75 mg/d C for 6 mo + P 3 mo</td>
<td>No symptoms at 6 mo</td>
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<td>240</td>
<td>20</td>
<td>15 d after LTx</td>
<td>⊗ 10 × 19</td>
<td>150 mg/d A + 75 mg/d C for 6 mo + Ac 3 mo</td>
<td>No symptoms at 20 d</td>
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<tr>
<td>Loyalka et al. (2012)</td>
<td>Information</td>
<td>8</td>
<td>Information NA</td>
<td>⊗ 10 × 25</td>
<td>325 mg/d A, 75 mg/d C</td>
<td>No symptoms at 1 mo</td>
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<td>Mohamed Mydlin et al. (2012)</td>
<td>Information</td>
<td>7</td>
<td>No stent, only dilatation</td>
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<td>Information NA</td>
<td>Recurrence after 2 mo, died 6 wk later</td>
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<td>&gt;1 y after LTx</td>
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<td>A + C + W for 1 mo, after A + W</td>
<td>No symptoms at 3 mo</td>
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<td>Information</td>
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<td>LMWH + W for 20 d, after W + C</td>
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<td>Current study, case 1</td>
<td>147</td>
<td>9</td>
<td>131 d after LTx</td>
<td>⊗ 12 × 26</td>
<td>A 80 mg/d</td>
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<td>Current study, case 2</td>
<td>172</td>
<td>12</td>
<td>10 d after LTx</td>
<td>⊗ 10 × 20</td>
<td>LMWH 60 mg, after Ed 30 mg</td>
<td>No symptoms at 4 mo</td>
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A, acetylsalicylic acid; Ac, acenocoumarol; ANG, angiography; BLL, bilateral lobar lung; C, Clopidogrel; CLPV, common left pulmonary vein; COPD, chronic obstructive pulmonary disease; CTA, computed tomography angiography; Ed, edoxaban; F, female; FA, fibrosing alveolitis associated with rheumatoid arthritis; GIT, gastrointestinal tract; HP, hypersensitivity pneumonitis; IP, interstitial pneumonitis; IPF, idiopathic pulmonary fibrosis; IPH, idiopathic pulmonary hypertension; LLPV, lower left pulmonary vein; LMWH, low-molecular-weight heparin; LPV, left pulmonary vein; LSL, left single lung; LTx, lung transplantation; M, male; NA, not available; P, phenprocoumon; PG, pressure gradient; PPCFV, peak pulmonary cuff velocity; PV, pulmonary vein; PVA, pulmonary vein surface area; PVO, pulmonary vein occlusion; PVS, pulmonary vein stenosis; RF, respiratory failure; RSL, right single lung; SSA, stenotic segment area; SSL, sequential single lung; TE, transesophageal echocardiography; TX, transplantation; ULPV, upper left pulmonary vein; URPV, upper right pulmonary vein; W, warfarin; ⊗, diameter.

*Clinical presentation: 1, respiratory insufficiency; 2, pulmonary edema; 3, pleural effusions.
watchful waiting) has been described in patients with mild PVS after atrial fibrillation ablation. It can lead to symptom improvement due to collateral formation or recruitment.² In the case of PVO, immediate treatment is required.

Surgical revision after transplantation can be challenging because of adhesions and the risk of graft loss, requiring retransplantation. An endovascular approach offers a valuable alternative with limited risk of complications, including LA perforation, PV dissection, stent embolization, or thrombosis.³ However, endovascular balloon dilatation of a fresh anastomosis can lead to rupturing and uncontrollable bleeding. In our experience, the best approach is interrogation with a soft compliant balloon (20% larger than the surrounding vessel) to assess the stretchability of the anastomosis, followed by stent implantation at the assessed size, which can be further dilated at a later stage.

In conclusion, transatrial endovascular treatment of PVS/PVO after LTx seems an effective and safe treatment that should be considered in the surgical LTx armamentarium and executed carefully to tackle these challenging complications.

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Disclosure

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Data availability

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ORCID

Michaela Orlitová https://orcid.org/0000-0002-5295-9315
Marc Gewillig https://orcid.org/0000-0002-4595-5922
Jan Van Slambrouck https://orcid.org/0000-0002-7069-1535
Dirk Vlasselaers https://orcid.org/0000-0002-9234-3419
Bart Jacobs https://orcid.org/0000-0002-1218-157X
Arne P. Neyrinck https://orcid.org/0000-0001-9930-8045
Lieven Depypere https://orcid.org/0000-0001-8230-5649
Laurent Godinas https://orcid.org/0000-0003-2214-5879
Robin Vos https://orcid.org/0000-0002-3468-9251
Geert M. Verleden https://orcid.org/0000-0003-3048-2429
Dirk E. Van Raemdonck https://orcid.org/0000-0003-1261-0992
Lauren J. Ceulemans https://orcid.org/0000-0002-4261-7100

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