



Case Report

Traumatic Pulmonary Hypertension Secondary to Arteriovenous Fistula and Remote Gunshot Wound

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ABSTRACT

Pulmonary hypertension is a known complication of high-flow arteriovenous fistulas (AVFs). We present a case of a 58-year-old man who sustained a gunshot wound 6.5 years before presentation for worsening pulmonary hypertension (PH). After diagnostic workup, the PH was attributed to a gunshot-related AVF. Exercise capacity and echocardiographic parameters improved after successful ligation of the AVF. This case highlights a rare and correctable cause of PH that requires careful investigation and multidisciplinary expertise for treatment.

RÉSUMÉ

L'hypertension pulmonaire est une complication connue des fistules artérioveineuses (FAV) à haut débit. Nous présentons le cas d'un homme de 58 ans qui a subi une blessure par balle 6,5 ans avant l'aggravation de son hypertension pulmonaire. Après les analyses diagnostiques, l'hypertension pulmonaire a été imputée à une FAV liée à la blessure par balle. Une amélioration de la capacité d'exercice et des paramètres échocardiographiques a été observée une fois réussie la ligature de la FAV. Ce cas met en évidence une cause rare et corrigible d'hypertension pulmonaire qui nécessite une analyse attentive et dont le traitement requiert des compétences multidisciplinaires.

Arteriovenous fistula (AVF) is a known complication of penetrating trauma.¹ We present a patient with long-standing traumatic AVF eventually causing pulmonary hypertension (PH). Traumatic AVF is a rare etiology of PH that may resolve with surgical ligation.

Case Summary

A 58-year-old man presented to the hospital with a gunshot wound to the right buttock. His medical history included hypertension (treated with ramipril), obesity, a remote 60 pack-year smoking history, and chronic back pain. Imaging revealed 2 rectal perforations requiring emergent laparotomy with loop sigmoid colostomy. The largest piece of the rifle bullet was removed from the subcutaneous tissue of the left anterior thigh, although many small fragments remained in the pelvis (~50).

Contrast-enhanced computed tomography showed stranding near the left common femoral artery and vein, with suspicion of pseudoaneurysm. He was discharged 1 month later.

One year after his gunshot injury, the patient experienced subacute leg swelling, and ultrasound imaging of his leg showed evidence of an AVF. Angiography confirmed a large, complex fistula between his left profunda femoral artery and common femoral vein (Fig. 1). Vascular surgery was consulted. By balancing surgical risk with then mild symptoms, repair was deferred unless he became symptomatic or cardiac decompensation occurred.

The patient was referred to cardiology 6.5 years after his initial presentation (age 65 years) because of transthoracic echocardiogram (TTE) evidence of PH with a pulmonary artery systolic pressure (PASP) of 68 mm Hg (summary in Supplemental Table S1, Fig. 2 for representative images). Connective tissue disease workup, thyroid-stimulating hormone, B-type natriuretic peptide, liver transaminases, and liver ultrasound results were all within normal limits. Pulmonary function test results showed decreased expiratory reserve volume (0.14 L; ref. 1.2–1.9 L) and functional reserve capacity (2.13 L; ref. 2.3–5.2 L), thought to reflect his obesity (height 179 cm, weight 125 kg, body mass index 39 kg/m²).

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See page 229.e13 for disclosure information.

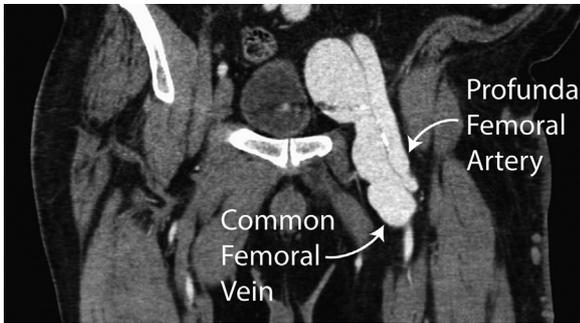


Figure 1. Coronal computed tomography angiography image demonstrating a large arteriovenous fistula (AVF) in the patient's left groin, with vessels labelled.

Polysomnography had been completed 4 years prior showing moderate obstructive sleep apnea. A ventilation-perfusion scan was not completed because of claustrophobia.

His PH was initially attributed to inadequately treated obstructive sleep apnea. Despite intermittent continuous positive airway pressure therapy, his estimated PASP

increased (Supplemental Table S1) and left leg swelling worsened. His functional status worsened to World Health Organization class II, which was attributed to the fistula. Left heart catheterization revealed mild, nonobstructive coronary artery disease. Right heart catheterization (RHC) demonstrated high cardiac index (4.9 L/min/m^2), high mean pulmonary artery pressure (63 mm Hg), high pulmonary artery systolic pressure (92 mm Hg), high pulmonary artery wedge pressure (28 mm Hg), no diastolic pressure gradient (diastolic pulmonary artery pressure – pulmonary artery wedge pressure; 0 mm Hg), and normal peripheral vascular resistance (2.4 Woods units) (Supplemental Table S2). These results were consistent with postcapillary PH due to left-sided diastolic cardiac dysfunction (Group 2 PH) and high-output heart failure (Group 5 PH).²

Vascular surgery was planned to ligate the patient's proximal left profunda fistula, leaving distal branches of the AVF patent because of massive venous dilation and concern for subsequent venous thromboembolism. The surgeon also prescribed a low-intensity exercise program. Repeated RHC immediately before surgery (age 66 years) showed worsening PH and increasing cardiac output (Supplemental Table S2).

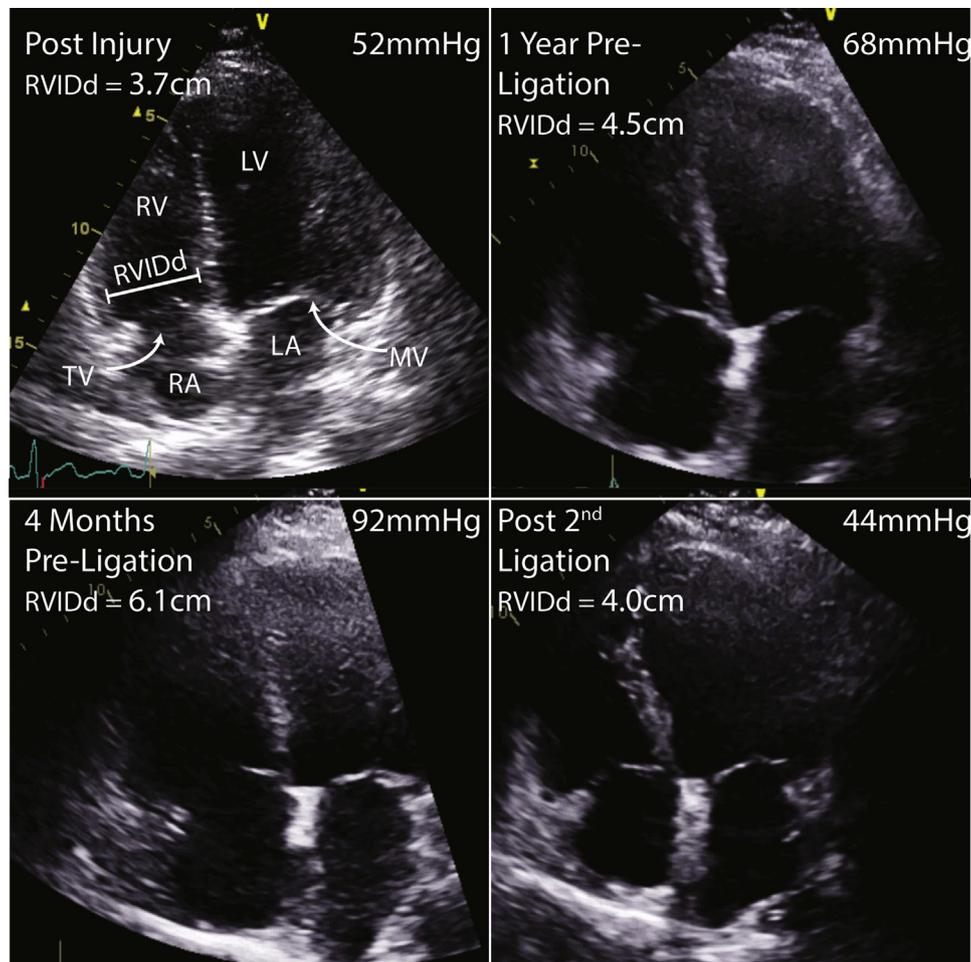


Figure 2. Echocardiogram images showing the evolution of the right ventricular internal diameter in diastole (normal < 4.2 cm). Relative time from surgery and right ventricular internal diameter in diastole indicated in the upper left of each image. Estimated pulmonary artery systolic pressure (PASP) (mm Hg) is indicated in the upper right of each image. LA, left atrium; LV, left ventricle; MV, mitral valve; RA, right atrium; RV, right ventricle; RVIDd, right ventricular internal diameter in diastole; TV, tricuspid valve.

Ligation was performed successfully, and low-dose apixaban was started as venous thromboembolism prophylaxis. Post-surgical RHC showed a reduction in cardiac output and small decrease in the mean pulmonary artery pressure (Supplemental Table S2). He recovered uneventfully and was followed in both the vascular surgery and PH clinics. Six-minute walk distance (6MWD) was 363 m (73% of predicted) 4 months after surgery.

His left leg swelling showed gradual improvement before subacute worsening 6 months after his repair. TTE showed improvement of PASP, although it remained elevated (Supplemental Table S1). Repeat 6MWD 8 months after surgery was essentially unchanged (347 m, 72% predicted). Repeat angiography (age 67 years) was performed, which revealed enlargement of the distal left profunda femoral artery fistula. A second surgery was performed to ligate the remaining distal branches of the fistula. Three months after definitive repair of the fistula, the patient's TTE showed further decrease in PASP (Supplemental Table S1), body mass index had decreased to 33.3 kg/m² with exercise, 6MWD improved to 448 m (87% of predicted), and functional class improved to World Health Organization class I.

Discussion

PH is a rare complication of traumatic AVF, and AVFs may rarely present decades after the inciting injury.¹ Given that high-flow AVFs may lead to high-output heart failure and PH, it is important for clinicians to recognize this potentially reversible etiology.³ Our patient showed severely elevated PASP, worsening RV dilation, and worsening left heart function that was found to be reversible with surgical ligation. This etiology may be initially obscure because of a potentially

long period between the initial injury and the eventual sequelae.

Conclusion

In patients with traumatic AVF who develop exercise limitation, with or without signs of right heart failure, clinicians should have a high index of suspicion for the development of PH and consider correction of the AVF.

Disclosures

The authors have no conflicts of interest to disclose.

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Supplementary Material

To access the supplementary material accompanying this article, visit the online version of the Canadian Journal of Cardiology at www.onlinecjc.ca and at <https://doi.org/10.1016/j.cjca.2018.12.003>.