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Case Report

Successful Treatment of a Stenotic Pulmonary Vein to Left Atrium Conduit With a Drug-Eluting Stent

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ABSTRACT

Partial anomaly of the pulmonary venous return is a rare congenital condition treated with surgical redirection of the blood flow through the creation of a conduit to the left atrium. We report the case of a stenotic pulmonary vein to left atrium conduit successfully treated with the implantation of a drug-eluting stent. Pulmonary vein or conduit stenosis is generally treated with balloon dilation or bare-metal stent but is often met with underwhelming outcomes. Given the successful outcome of the case presented, drug-eluting stents may represent an attractive treatment option in suitable anatomies.

RÉSUMÉ

Les anomalies partielles du retour veineux pulmonaire sont des anomalies congénitales rares traitées par une intervention chirurgicale visant à rediriger le flux sanguin par la création d'un conduit vers l'oreillette gauche. Nous rapportons un cas de veine pulmonaire sténosée au conduit de l'oreillette gauche pour laquelle l'implantation d'une endoprothèse médicamentée s'est avérée une réussite. La sténose de la veine pulmonaire ou du conduit est généralement traitée par la dilation par ballonnet ou l'implantation d'une endoprothèse non médicamentée nu, mais les résultats sont souvent décevants. Compte tenu des résultats favorables observés pour ce cas, les endoprothèses à élution de médicamentées peuvent constituer une option attrayante de traitement dans les cas où l'anatomie s'y prête.

Anomalous pulmonary venous connection (APVC) is a congenital defect defined by the abnormal venous return from at least 1 pulmonary vein to the right atrium or systemic vein. The physiological consequence is a left-to-right shunt with potential right-sided volume overload or pulmonary hypertension. We report the case of a patient who developed symptomatic stenosis of a pulmonary vein to left atrium (LA) conduit.

Clinical Case

A 45-year-old man presented with New York Heart Association class 3 and fatigue progressing over the last 6 months. At age 24 years, he had undergone surgical correction of an atrial septal defect with redirection of the anomalous right superior pulmonary vein to the LA through a conduit made of autologous pericardium and connected to the LA through the atrial septal defect patch.

Transesophageal echocardiography (TEE) demonstrated flow acceleration in the conduit near the LA anastomosis (1.5 m/sec). Multidetector computed tomography showed a severe stenosis of the conduit and collaterals from the right upper pulmonary lobe to the inferior lobe (Fig. 1). Baseline stress test result was clinically abnormal and limited to 7.5 metabolic equivalents.

Interventional cardiology and cardiac and thoracic surgery were consulted. The decision was made to proceed with percutaneous treatment.

Under general anaesthesia and TEE guidance, access to the LA was gained with a superiorly located transseptal puncture. Heparin was given to achieve an activated-clotting time greater than 250 seconds. A deflectable Agilis catheter (Abbott Inc, Santa Clara, CA) was advanced to the LA and deflected inferiorly.

A multipurpose guiding catheter was telescoped into the Agilis and maneuvered to approximate the conduit ostium, as guided by TEE (Video 1, view video online). The conduit was opacified and found nearly occluded in the segment proximal to the LA anastomosis. The length of

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See page 149 for disclosure information.

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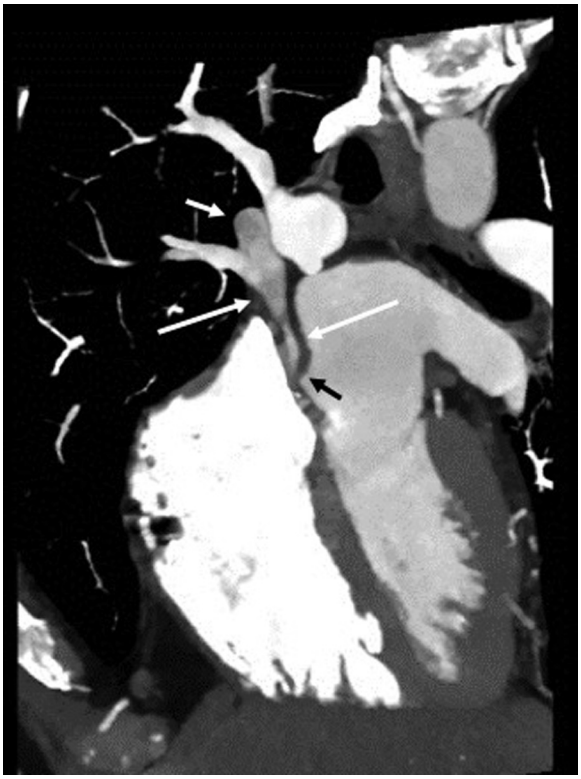

Figure 1. Multislice cardiac computed tomography. The surgical conduit (**long arrows**) connects the anomalous right superior pulmonary vein (**short white arrow**) to the left atrium (LA). The conduit is severely stenosed near its anastomosis to the LA (**black arrow**).

stenosis could not be accurately assessed with minimal contrast retrogradely opacifiying the conduit. A 0.014-inch coronary wire was advanced into the conduit, and predilatation was performed followed by repeat angiography for stent selection. Because preintervention TEE, multidetector computed tomography, and operative protocol did not provide the reference diameter, we used the largest angiographic diameter (3.5 mm) of the conduit as the reference and implanted a 4.0 × 15-mm drug-eluting stent (DES). With control angiography showing incomplete coverage, a second 4.0 × 15-mm stent was implanted; both were postdilated up to 5 mm. The angiographic result was excellent (Video 2, view video online), poststenting gradient across the stented area was 3 mm Hg, and collateral drainage disappeared (Fig. 2; Video 3, view video online). The patient was discharged the next day under dual antiplatelet therapy.

TEE was performed 2 months later showing normal flow and stable 3 mm Hg mean gradient across the stented area. The patient rapidly improved to New York Heart Association class I. Stress test performed 1 year later showed clinically negative results, and the exercise tolerance improved from 7.5 to 9.3 metabolic equivalents.

Discussion

Pulmonary vein stenosis is a rare condition. In the adult population, it follows pulmonary vein isolation in most cases but can also develop after surgical repair of APVC. Postrepair conduit stenosis has an incidence of 10% to 17% according to multiple series.[1-3] Although this complication usually occurs within 1 year after surgery, late stenosis is possible. In a recent review,[4] freedom from reintervention and restenosis at 5 years were 55% and 56%, respectively. When untreated, pulmonary vein stenosis leads to shortness of breath, pulmonary hypertension, and even death. In the case presented, symptoms developed late after surgery in a patient without regular follow-up. Although uncommon, this can be explained by the progression of exercise-induced pulmonary hypertension despite normal pulmonary pressure at rest or late stenosis of an unknown mechanism.

Balloon angioplasty of stenotic native pulmonary veins is met with frequent recoil and restenosis. Bare-metal stents improve results, but reintervention remains common, up to 46%.[5] Data suggest long-term patency is best achieved in veins treated with stents at least 9 to 10 mm in diameter. Whether these findings in native pulmonary veins apply to stenotic conduits is unknown. In our case, it seemed the conduit was smaller than a typical adult pulmonary vein.

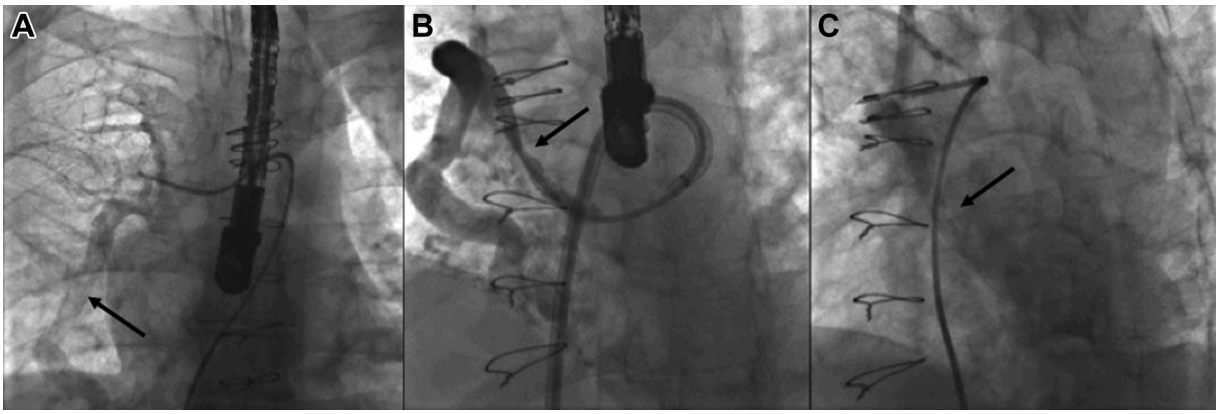

Figure 2. Procedural angiography. (**A**) Levophase of selective pulmonary artery angiography; blood flood draining through a large collateral (**arrow**) toward the right lower pulmonary vein. (**B**) Retrograde angiography of the conduit after the first stent implantation showing residual stenosis (**arrow**) upstream from the first stent. (**C**) Poststenting right upper pulmonary artery angiography: Venous return flows through the stented conduit (**arrow**) rather than collaterals.

Considering the likelihood of restenosis after pulmonary vein stenting and the well-documented ability of DES to prevent intimal hyperplasia in coronary arteries, DES may represent an attractive option when applicable. In a small cohort, De Potter et al.[6] described a restenosis rate of only 14% at 3-month follow-up after DES (mean diameter 4.3 mm) implantation for the treatment of stenosis complicating pulmonary vein isolation.[6] In our case, the conduit diameter seemed in the range of a postdilated large coronary stent. The small gradient obtained after stenting may represent underexpansion of the conduit or a gradient inherent to the conduit itself. Despite this, DES ability to prevent restenosis could yield a better outcome than a larger, restenosed bare-metal stent.

To our knowledge, this is the first report of pulmonary vein to LA conduit DES implantation in an adult patient. Initial results and midterm evolution are promising. Given the successful outcome of the case presented and underwhelming midterm results of conventional treatment of pulmonary vein or pulmonary vein to LA conduit stenosis, a DES may represent an attractive treatment option when the anatomy is suitable.

Conclusion

Pulmonary vein stenosis after surgical correction of partial APVC is a rare condition. Given the excellent outcome of this patient, DES implantation in such conduits or relatively small pulmonary veins may be an attractive alternative.

Disclosures

The authors have no conflicts of interest to disclose.

References

1. Lamb RK, Qureshi SA, Wilkinson JL, et al. Total anomalous pulmonary venous drainage. Seventeen-year surgical experience. J Thorac Cardiovasc Surg 1988;96:368-75.

2. Sano S, Brawn WJ, Mee RB. Total anomalous pulmonary venous drainage. J Thorac Cardiovasc Surg 1989;97:886-92.

3. Van de Wal HJ, Hamilton DI, Godman MJ, et al. Pulmonary venous obstruction following correction for total anomalous pulmonary venous drainage: a challenge. Eur J Cardiothorac Surg 1992;6:545-9.

4. Kalfa D, Belli E, Bacha E, et al. Outcomes and prognostic factors for postsurgical pulmonary vein stenosis in the current era. J Thorac Cardiovasc Surg 2018;156:278-86.

5. Prieto LR, Schoenhagen P, Arruda MJ, Natale A, Worley SE. Comparison of stent versus balloon angioplasty for pulmonary vein stenosis complicating pulmonary vein isolation. J Cardiovasc Electrophysiol 2008;19:673-8.

6. De Potter TJ, Schmidt B, Chun KR, et al. Drug-eluting stents for the treatment of pulmonary vein stenosis after atrial fibrillation ablation. Europace 2011;13:57-61.

Supplementary Material

To access the supplementary material accompanying this article, visit *CJC Open* at https://www.cjcopen.ca/ and at https://doi.org/10.1016/j.cjco.2019.02.004.