Partial anomalous venous connection with intrapulmonary dual drainage: Transcatheter treatment of a rare entity

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ABSTRACT

Partial anomalous pulmonary venous return (PAPVR) is a rare congenital condition, and dual-drainage connection PAPVR to the left atrium has been reported in a few cases in the literature; in which cases, percutaneous catheterization was successfully used in lieu of surgery. We, hereby, describe a 7-month-old boy with a functional single-ventricle physiology with dual drainage of the left upper pulmonary vein to the left atrium and the innominate vein. Appropriate recognition of this entity allowed safe occlusion of the anomalous draining vein.

Keywords: Cardiac catheterization, congenital heart disease, dual pulmonary venous drainage, partial anomalous venous connection

A 7-month-old boy with a functional single-ventricle physiology, who underwent a bidirectional cavopulmonary anastomosis, had persistent hypoxemia and was referred for cardiac catheterization. His mean Glenn pressure was 13 mmHg with a pulmonary artery oxygen saturation of 55%. His systemic saturations were low for the Glenn physiology (75%) with an average pulmonary vein saturation of 78%. There was 1 mmHg gradient from the superior vena cava to the left pulmonary artery due to proximal left pulmonary artery stenosis. He had a normal systemic cardiac index (4.18 L/min/m²) with a pulmonary-to-systemic blood flow ratio of 0.55. His calculated pulmonary vascular resistance was 2.18 iWU. PA angiogram demonstrated reduced filling of the left upper lobe, and selective left upper lobe angiogram demonstrated an anomalous left upper pulmonary vein connecting to the left innominate vein through a vertical vein (VV) [Video 1]. Selective contrast injection in the VV demonstrated

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intrapulmonary dual drainage of the left upper lobe [Figure 1 and Video 2]. This innominate vein (mean pressure of 13 mmHg) to the anomalous left upper lobar pulmonary vein (mean pressure of 8 mmHg) connection led to right-to-left shunting and was responsible for the systemic desaturation in this patient. Due to this right-to-left shunting from the innominate vein to the left upper lobe, there is a competing flow to the left upper lobe. This was appreciated as reduced filling of the left upper lobe on pulmonary artery angiogram due to the unopacified blood from the left innominate vein anomalous connection. Due to the dual drainage, the VV was occluded using a 5Q Medtronic microvascular plug [Figure 2 and Video 3]. Postocclusion, the hypoxemia improved with a saturation of ~ 85%. In addition, the patient also had left pulmonary artery stenosis. This was relieved using an 8 mm \times 29 mm GORE VBX® Balloon Expandable Stent.

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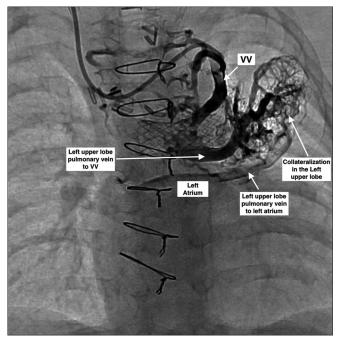


Figure 1: Selective contrast injection in the VV demonstrated intraparenchymal duplicate drainage of the left upper lobe. VV: Vertical vein

Partial anomalous pulmonary venous return (PAPVR) is a rare congenital condition, and dual-drainage connection PAPVR to the left atrium has been reported in a few cases in the literature; in which cases, percutaneous catheterization was successfully used in lieu of surgery.^[1] Patel et al. described anatomy, distinctly noting the "extrapulmonary" and "intrapulmonary" separation of the dual drainage.^[2] Here, we report a case of intraparenchymal duplicate drainage in a patient with a complex cardiac history, who underwent a successful transcatheter occlusion and improvement in systemic hypoxemia. Even with modern imaging modalities, this entity may go undetected; hence, a high index of suspicion is recommended in appropriate clinical circumstances. Recognition of this entity in angiography can allow prompt interventional treatment which has the potential to shorten the length of stay and avoid repeat surgical intervention.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the

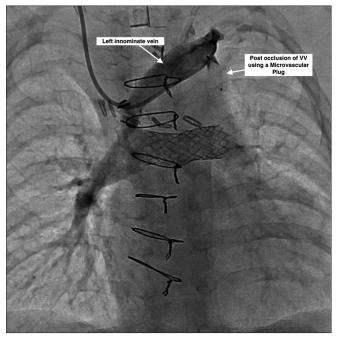


Figure 2: Contrast injection in the innominate vein after occlusion of the VV using a microvascular plug. VV: Vertical vein

patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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