CASE REPORT

Anomalous Unilateral Single Pulmonary Vein — a Rare Mimicker of Pulmonary Arteriovenous Malformation

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ABSTRACT
Anomalous unilateral single pulmonary vein is a rare congenital anomaly that can masquerade as a pulmonary arteriovenous malformation on computed tomography scan. This report describes a woman with cough for whom an initial diagnosis of pulmonary arteriovenous malformation was made based on plain X-ray and computed tomography scan. Anomalous unilateral single pulmonary vein was diagnosed by selective pulmonary angiogram that aimed for embolisation of the arteriovenous malformation. Since anomalous unilateral single pulmonary vein can have a non-specific clinical appearance, an abnormal pathologic vascular shadow on a chest X-ray should raise the suspicion of a congenital pulmonary vascular abnormality and initiate further imaging if indicated.

Key Words: Congenital; Angiography; Arteriovenous malformations; Computed tomography; Pulmonary veins

INTRODUCTION
Anomalous unilateral single pulmonary vein (AUSPV) is a rare anomaly of the pulmonary venous system. AUSPV with a normal connection to the left atrium is exceedingly rare. Differentiating this rare anomaly from other conditions is important because isolated AUSPV does not require treatment, so thoracic surgeons should be aware of this anomaly before performing lung resection. This report describes a patient with AUSPV mimicking pulmonary arteriovenous malformation (AVM), with a definitive diagnosis established with the help of pulmonary angiogram.

CASE REPORT
A 50-year-old woman was referred to the Department of Radiology and Imaging, Queen Elizabeth Hospital, Hong Kong, from the Respiratory Medical Department after several episodes of cough in 2001. The initial chest X-ray (Figure 1) and thoracic computed tomography (CT) scan performed at a private hospital showed a tubular vascular malformation in the left lung (Figure 2), suggestive of a pulmonary AVM. Only a hard copy of the axial plane CT images of 7.5-mm slice thickness was provided for reference. The patient had no history of dyspnoea, bleeding, or clinical signs of Rendu-Osler-Weber disease.

Selective pulmonary angiography intended for embolisation of the pulmonary AVM was performed, and showed normal arterial anatomy. The venous phase displayed a single dilated tortuous vein forming a loop that extended from the left upper lobe to the left lower lobe, draining the entire left lung into the left atrium (Figures 3a and 3b). The diagnosis of an AUSPV was made. The vasculature in the right lung was normal. Embolisation was not performed due to the absence of an AVM.

DISCUSSION
AUSPV is a rare congenital anomaly. Single pulmonary veins can occur in association with other vascular or pulmonary malformations such as a bilateral AUSPV, a contralateral partial anomalous venous connection, and malformations of the bronchopulmonary airways and the related arterial blood supply. Confusion with other conditions such as scimitar syndrome, partial anomalous pulmonary venous return, pulmonary varices or venous aneurysms, pulmonary AVM, and parenchymal pulmonary masses has been reported.1-6 The anomalous
single pulmonary vein drains appropriately into the left atrium. Although extremely rare, this venous anomaly is important to recognise because it requires no further therapeutic intervention.

Usually, AUSPV is an isolated and asymptomatic condition that is diagnosed accidentally. Therefore, an abnormal pathologic vascular shadow on a chest X-ray, even in the absence of arterial blood gas and haemoglobin abnormalities, should raise the suspicion of a congenital pulmonary vascular abnormality and initiate further imaging if indicated.

Selective pulmonary angiography, the modality of choice for revealing the exact fistula site in pulmonary AVM, is diagnostic of this unusual abnormality. Subsequent embolisation of the lesion could then be carried out in the same setting, as was initially planned for this patient. As selective pulmonary angiography is considered an invasive procedure, multidetector row CT (MDCT) with intravenous contrast has replaced it as the method of choice for diagnosing AUSPV. With the MDCT scanner, visualisation of the AVM and its nidus, venous drainage to the left atrium of the heart, and its origin from the pulmonary artery can be demonstrated superbly by means of different post-processing techniques such as 3-dimensional volume rendering, multiplanar reconstruction, and curve-reformation techniques. The advantages of MDCT are that it is considered as a non-invasive, fast, and effective modality for diagnosing vascular abnormality. For those centres with no provision for an MDCT scanner, it is important to alert the clinician not to perform any biopsy or surgical resection before the differentiation between vascular abnormality and neoplastic mass can be made. Even with a simple CT scanner, one can tell whether the lesion is vascular in nature by detailed density measurement and comparison of the density between the lesion and the adjacent pulmonary venous or arterial vasculature. MDCT with intravenous contrast should be recommended for the differential diagnosis of any

Figure 1. (a) Chest X-ray and (b) its magnified view showing a tubular structure in the left lower zone.

Figure 2. Contrast-enhanced computed tomography scan showing a tubular vascular malformation in the left lower lobe.
Anomalous Unilateral Single Pulmonary Vein

Figure 3. The venous phase of the pulmonary angiogram displaying a single dilated tortuous vein forming a loop that extended from the left upper lobe to the left lower lobe, draining the entire left lung into the left atrium.

pulmonary vascular malformation if vascular intervention or surgery is contemplated.

REFERENCES