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An Occult Congenital Fistula Between the Descending Aorta and the Left Pulmonary Vein in an Adult Presenting With Recurrent Episodes of Hemoptysis

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We report a case of recurrent hemoptysis due to an occult congenital fistula between the descending aorta and the left pulmonary vein in a 25-year-old female patient. The anomaly was confirmed by contrast-enhanced CT scan and angiography. No abnormality was noted in the bronchia and pulmonary arteries. The patient was successfully managed by simple ligation of the fistula. To our knowledge, this is the first reported case of adult-onset hemoptysis caused by an occult congenital fistula between the descending aorta and a pulmonary vein.

CHEST 2013; 143(2):549–553

Abbreviations: DAPVF = descending aorta-pulmonary vein fistula

Hemoptysis can be caused by a variety of conditions, including tracheobronchial diseases, pulmonary parenchymal lesions, primary vascular anomalies, and systemic coagulopathies.[1] Here, we present the first report, to our knowledge, of an adult case of hemoptysis due to congenital descending aorta-pulmonary vein fistula (DAPVF).

Case Report

A 25-year-old female teacher was admitted to our hospital because of recurrent hemoptysis over 1 year. Initially the patient complained about expectoration of blood-streaked mucus, which occurred several times over 6 months. The

Manuscript received December 20, 2011; revision accepted June 1, 2012.
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DOI: 10.1378/chest.11-3229

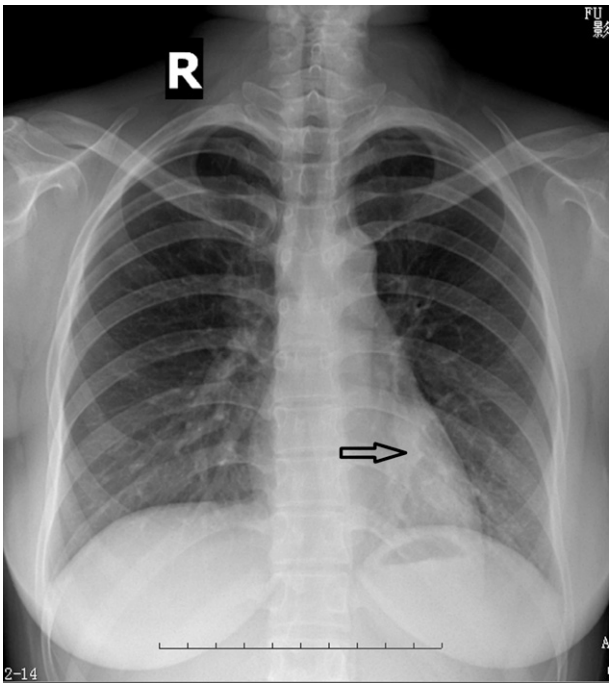

FIGURE 1. Preoperative chest radiograph shows a smooth and highly dense opacity in the left lower lobe behind the heart (black arrow).

situation worsened in the subsequent month, and the patient reported one to three expectorations of a scant volume of bright red blood (about 5 mL) or blood clot. Exercise or menstruation did not appear to have any apparent effect on these episodes. The patient denied having fever, chest pain, night sweats, shortness of breath, or any anticoagulant use. On physical examination, she appeared to be in good general condition.

A left parasternal grade 3/6 systolic murmur was heard over the fourth intercostal space, with a normal pulmonary valve closure (P2) sound. Echocardiography showed a normal cardiac contour without any defect or anomalous communication. Chest radiograph (Fig 1) and fluoroscopy (Video 1) revealed a smooth, highly dense opacity in the left lower lobe. Contrast-enhanced CT scan of the chest showed an anomalous arterial branch, 9 mm in the narrowest inner diameter, that arose from the descending aorta and was connected with the left pulmonary vein before its entry into the left atrium (Figs 2A-D). The pulmonary vein was dilated; the bronchial tree (Fig 2E) and pulmonary arterial branches (Fig 2F) appeared normal. Angiography showed that the contrast agent entered the lower tributaries of the left inferior pulmonary vein through the DAPVF (Fig 3A) before returning to the left inferior pulmonary vein and joining the left atrium (Fig 3B, Videos 2, 3).

Proliferating vessels suffused the basal segments of the left inferior lobe obviously (Fig 4A). An anomalous large vessel originated from the descending aorta and then entered the left inferior lobe (Figs 4B, 4C). After ligation of the anomalous vessel through a left thoracotomy using the same surgical procedure as for patent ductus arteriosus, the aneurysmatic part of the anomalous vessel in the lung disappeared (Fig 4D). Intraoperative biopsy was not performed because the patient refused to provide informed consent.

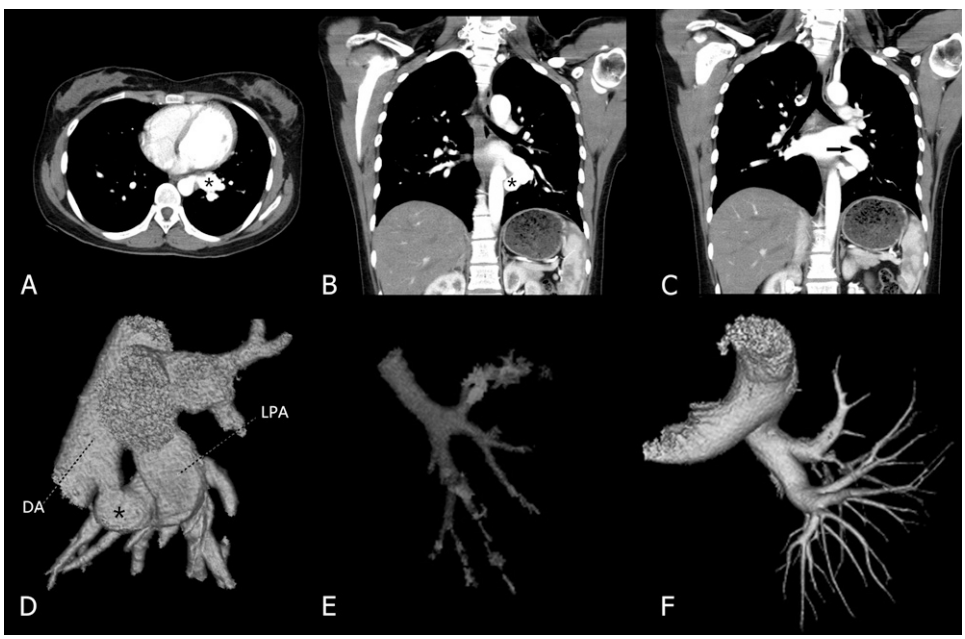

FIGURE 2. Contrast-enhanced CT scan shows the anatomic structure of DAPVF, the left bronchial tree, and left pulmonary branches. A, The anomalous systemic artery (*) arises from the descending aorta. B, The anomalous systemic artery (*) is connected with the left inferior pulmonary vein. C, The dilated inferior pulmonary vein (black arrow) joins the left atrium. D, Three-dimensional reconstruction of DAPVF. The narrowest inner diameter of DAPVF is 9 mm. E, The normal left bronchial tree. F, The normal left pulmonary arterial branches. DA = descending aorta; DAPVF = descending aorta-pulmonary vein fistula; LPA = left pulmonary artery.