

# Massive hemoptysis from a bronchial Dieulafoy lesion

A Case Report

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## Abstract

A 30 year old male with a history of recurrent pulmonary emboli presented with hemoptysis 6 weeks after discontinuing anticoagulation therapy. He was found to have submassive pulmonary embolism with elevated pulmonary artery pressures and underwent bilateral catheter-directed pharmacomechanical thrombolysis. On day 3 of admission he developed massive hemoptysis and bronchoscopy revealed a Dieulafoy lesion on the left upper lobe bronchial mucosa to which electrocautery and cryotherapy was applied. Bronchial arteriogram revealed dilated bronchial arteries and left bronchial artery embolization was performed after which he was started on sildenafil. His hemoptysis resolved and he was discharged home on warfarin.

## Case

A 30 year old African-American male with a history of two prior episodes of submassive and massive pulmonary embolism presented to the emergency department with acute hemoptysis. In 2005, the patient had a pulmonary embolism after a motor vehicle accident, and was treated with 6 months of anticoagulation. In 2007 he had a recurrent unprovoked massive pulmonary embolism requiring urgent surgical embolectomy and inferior vena cava filter placement. He had been maintained on anticoagulation with warfarin until 6 weeks prior to presentation, when he ran out of medication. The patient was an avid soccer player and reported not being able to participate in his normal level of play during the four weeks prior to admission.

On the day of presentation, the patient had hemoptysis which awoke him from sleep prompting him to seek medical attention. The patient's vital signs at presentation included a blood pressure 118/70, heart rate 70, respiratory rate 18, and oxygen saturation 97% on room air. Physical examination was significant for the absence of jugular venous distention, clear lungs, normal body habitus, regular heart rate without gallop or murmur. A right ventricular heave was present. Initial laboratory values included a troponin < 0.05 mg/dL and B-type natriuretic peptide < 20 pg/dL. In the emergency department, a Computed Tomography (CT) scan with intravenous contrast revealed a large right sided main pulmonary artery embolus with chronic pulmonary vascular changes noted in the left main pulmonary artery and near-complete occlusion of the left lower lobe segmental pulmonary artery. Echocardiogram revealed normal left ventricular systolic function with abnormal septal motion consistent with right ventricular pressure overload and estimated right ventricular systolic pressure of 56-61 mmHg. CT scan of the abdomen revealed a nonocclusive thrombus within the inferior vena cava extending superior to the level of the inferior vena cava filter. Lower extremity ultrasound showed large

acute and chronic thrombus within the right external iliac vein extending into the proximal femoral vein.

Due to the extensive clot burden and finding of elevated right ventricular pressure, the patient underwent bilateral catheter-mediated pharmacomechanical thrombolysis (EKOS EkoSonic® Endovascular system; EKOS Corporation, Bothell, WA, USA) with catheter-directed infusion of tissue plasminogen activator at 1mg/hour for 24 hours.

The left and right mean pulmonary artery pressures were 42 mm Hg and 46 mm Hg respectively prior to the procedure and 31mm Hg and 46 mm Hg respectively at the conclusion of the procedure. Three hours after discontinuation of the infusion, at which time his coagulation studies were within normal limits, the patient developed recurrent bouts of hemoptysis totaling 240 mL over a 12 hour period. Bronchoscopy was performed which revealed an erythematous, vascular-appearing lesion on the left upper lobe bronchial mucosa (Figure 1).

### Cooper bronchoscopy 1

Image not found

<http://journal.pulmccm.org/wp-content/uploads/2015/06/Owens-bronchoscopy-11.png>

## Fig. 1: Fiber-optic bronchoscopy.

Top: Dieulafoy lesion (arrows) at the left upper lobe origin. Bottom: Dieulafoy lesion after electrocautery.

The lesion was not bleeding on initial viewing; however, gentle probing of the lesion induced approximately 30 cc of frank bright red blood. Hemostasis was initially achieved using bronchoscopic electrocautery; however, the patient continued to experience hemoptysis over the next several hours and subsequently underwent left bronchial artery embolization of a tortuous, dilated, ectatic left bronchial artery believed to be the culprit lesion (Figure 2).

### Cooper 2

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<http://journal.pulmccm.org/wp-content/uploads/2015/06/Cooper-2.png>

## Fig. 2: Bronchial angiography.

Left: dilated, tortuous, ectatic left bronchial artery. Right: left bronchial artery after successful embolization.

During this hospitalization the patient continued to experience episodes of hemoptysis for which subsequent bronchoscopies were again performed utilizing the instillation of epinephrine, electrocautery, and cryotherapy to achieve hemostasis. Following the use of cryotherapy, the patient did not experience recurrent hemoptysis and remained hemodynamically stable. He was started on sildenafil for pulmonary arterial hypertension and resumed warfarin without recurrent episodes of bleeding noted during outpatient follow up.

## Discussion

The lung has a dual blood supply. The pulmonary arteries originate from the right ventricle and supply blood to

the lung parenchyma. The bronchial arteries originate from the aorta and supply the walls of the conducting airways. These vessels form anastomoses near the alveolar ducts.<sup>1</sup> When the pulmonary circulation is compromised in circumstances such as chronic thromboembolic occlusion of the pulmonary vasculature, the bronchial arteries, which are capable of angiogenesis, hypertrophy and develop new bronchopulmonary anastomoses distal to the occluded pulmonary arteries. In one report of 79 patients with chronic thromboembolic pulmonary hypertension, there was a 6% incidence of hemoptysis.<sup>2</sup> This patient's chronic thromboembolic pulmonary hypertension resulted in compensatory bronchial artery dilatation and neovascularization, resulting in the formation of a bronchial Dieulafoy lesion and massive hemoptysis.

A Dieulafoy lesion is characterized by submucosal arterial dysplasia and is most commonly associated with gastrointestinal manifestations, however, several case reports document such lesions on the bronchial walls.<sup>3</sup> Most commonly they can be visualized on bronchoscopy as a small nodular lesion, typically enclosed with a white cap with a relatively normal overlying mucosa.<sup>4</sup> The culprit lesion most commonly presents itself in the context of hemoptysis which is why a Dieulafoy lesion should be considered in any patient with unexplained hemoptysis, particularly if contained to a single lobe.<sup>5</sup> The preferred treatment for Dieulafoy lesions include angiography with embolization, however their occurrence and optimal treatment have not been comprehensively studied. Success rates with embolization have been reported to be approximately 40% when a Dieulafoy lesion is suspected to be the underlying cause of hemoptysis.<sup>6</sup>

In this case, multimodality treatment was necessary to treat the massive hemoptysis which occurred due to bronchial artery hypertrophy and the formation of a bronchial Dieulafoy lesion. This was comprised of decreasing the pulmonary arterial pressure through pharmacomechanical clot disruption with the EKOS catheter and initiation of sildenafil, as well as direct treatment of the culprit portion of the bronchial circulation through both bronchial artery embolization and direct electrocautery and cryotherapy to the bleeding mucosal lesion.

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