

# Embolization of pulmonary artery aneurysms in a patient with Behçet's disease complicated by coil erosion into the airway

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## ABSTRACT

Behçet's disease is an inflammatory vasculitis with the unique feature of pulmonary artery aneurysms. We describe a patient with Behçet's disease and pulmonary artery aneurysms who presented with massive hemoptysis treated by coil embolization. Although there was immediate resolution of hemoptysis and improvement in hemodynamic status, 2 months later the patient reported a refractory cough and feeling of foreign body in her throat. Imaging demonstrated partial coil migration into the bronchus and trachea. Although endovascular intervention is the first-line treatment for massive hemoptysis, in patients with Behçet's disease, active inflammation and chronic steroid use may increase the risk of coil erosion and migration. (*J Vasc Surg Cases Innov Tech* 2022;8:193-5.)

**Keywords:** Behçet's disease; Pulmonary artery aneurysm; Coil migration

Behçet's disease, also known as oculo-orogenital syndrome, is an idiopathic inflammatory vasculitis whose distinguishing features include uveitis and oral and genital ulcers.<sup>1,2</sup> Pulmonary artery aneurysm (PAA) formation is a unique feature of Behçet's disease. PAAs in patients with Behçet's disease often manifest with hemoptysis, and are a leading cause of death in this patient population.<sup>2,3</sup> Endovascular embolization is the first-line treatment in such cases with few complications reported in the literature.<sup>4</sup>

## CASE PRESENTATION

The patient discussed in this case provided written informed consent, giving their permission to publish this case. The patient is 45-year-old woman with a history of Behçet's disease with pulmonary involvement including PAAs, factor V Leiden, and prior cerebral venous thrombosis and pulmonary embolism requiring lifelong anticoagulation who presented with massive hemoptysis. She initially underwent endovascular pulmonary artery coil embolization of two pulmonary aneurysms in the right lung (Fig 1). Embolization improved hemodynamic status (preprocedure blood pressure of 91/61 mm Hg, postembolization blood

pressure of 108/76 mm Hg), and resolution of hemoptysis. One month after coil embolization, the patient returned with massive hemoptysis which resolved after holding her anticoagulation. A chest radiograph was obtained; however, no cross-sectional imaging was performed. There was concern for ongoing active vasculitis owing to an elevated C-reactive protein and erythrocyte sedimentation, and her daily dose of steroids was increased from 10 mg to 40 mg.

Two months after embolization, the patient reported refractory cough and a foreign body sensation in her airway. A chest radiograph followed by a computed tomography angiogram were obtained for further evaluation. Partial coil migration from the right upper lobe aneurysm into the right main stem bronchus and trachea was seen (Fig 2).

The patient then was admitted to the hospital and a bronchoscopy was performed to evaluate and remove portions of the right upper lobe coil that had eroded into the airway (Fig 3). The patient's steroids were tapered and she ultimately underwent right upper lobectomy to remove the partially migrated coils in the anterior segment of the right upper lobe; the coils in the superior segment of the right lower lobe were left in situ. Surgical pathology demonstrated the known coil-filled PAA as well as vasculitis involving small to medium caliber vessels. Additionally, acute-on-chronic bronchitis, inflamed granulation tissue associated with the fistulized coil, and an adjacent aspergilloma were identified. The patient was discharged from the hospital 1 week after surgical resection, and remained in good condition without an episode of recurrent hemoptysis 6 months after the lobectomy (Fig 4).

## DISCUSSION

Endovascular intervention is a first-line lifesaving treatment for patients with Behçet's disease with massive hemoptysis owing to PAAs. A case series of 17 patients with Behçet's disease found that embolization of PAAs

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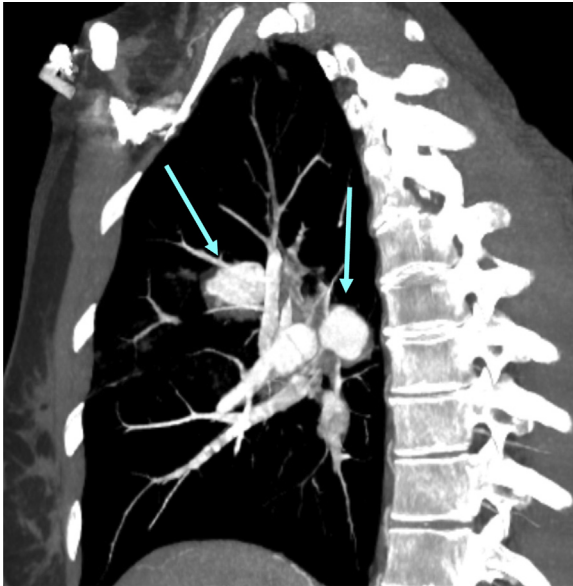
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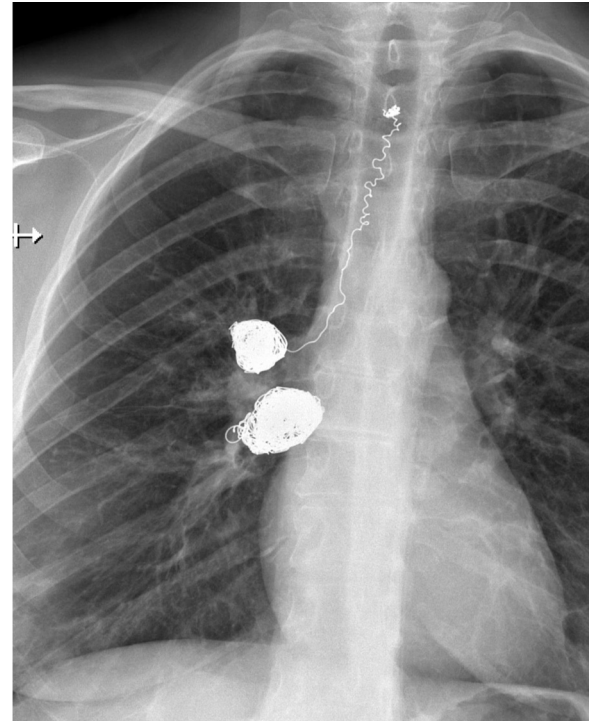
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**Fig 1.** Sagittal maximum intensity projection computed tomography angiography of the chest revealed two pulmonary artery aneurysms (PAAs) in the right lung (blue arrows).

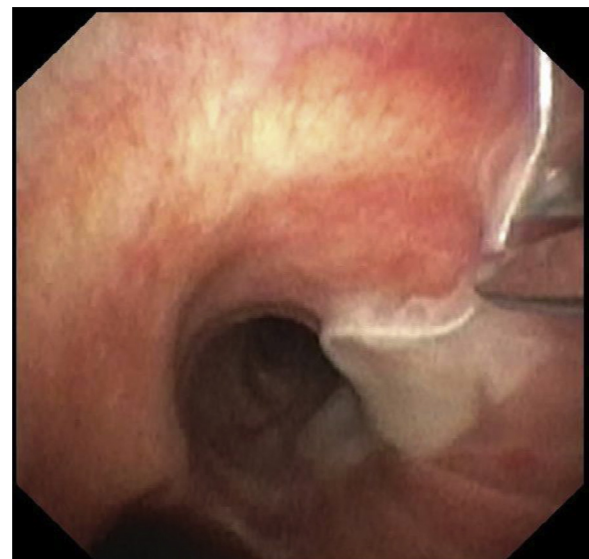


**Fig 2.** Radiographs of the chest were obtained owing to ongoing cough and complaint of a foreign body sensation. The posteroanterior radiograph shows two coil packs, the more superior coils in the anterior segment of the right upper lobe and the more inferior coils in the superior segment of the right lower lobe. There is cranial migration of coils from the anterior segmental artery of the right upper lobe into the adjacent bronchus.

was successful in achieving immediate control of hemoptysis in all patients.<sup>5</sup> Hemoptysis recurred in 41% of patients within 12 months, and one major complication was reported (acute exacerbation of preexisting pulmonary hypertension).<sup>5</sup> Coil erosion and migration into the mainstem bronchus is a rare complication of coil embolization with an unknown incidence, described in a case report occurring 2 years after embolization for which a left lower lobectomy with resection of the PAA and coil was performed.<sup>6</sup> Our case differs from this previously published case report given the short time between embolization and symptom onset. Bleeding risk after bronchoscopy to remove migrated endovascular coils is not known, but 1 series of 1035 bronchoscopies in children to remove aspirated foreign bodies reported only 6 cases of bleeding from the tracheobronchial system.<sup>7</sup>

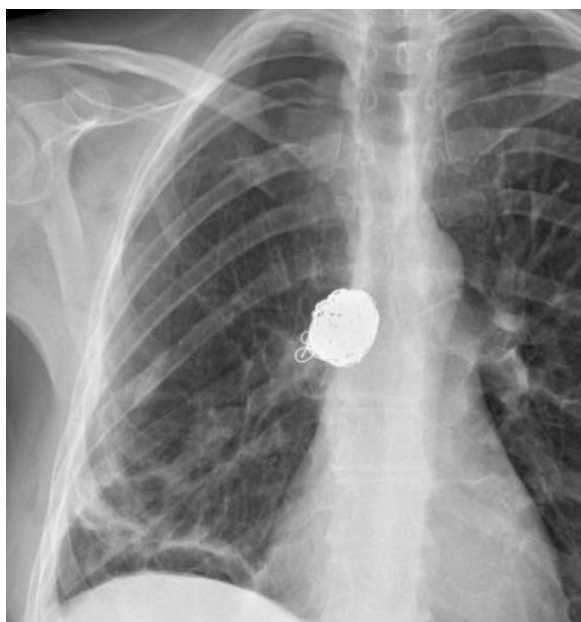
The first-line treatment for vascular Behçet's disease is immunosuppressive therapy.<sup>8</sup> Patients who present with massive hemoptysis and have failed medical therapy can be treated with endovascular intervention.<sup>8</sup> Surgery is reserved for patients who fail medical and/or endovascular treatment, or have a complication related to endovascular intervention.<sup>8</sup>

We hypothesize that chronic steroid use, ongoing active inflammation, and thin pulmonary artery walls led to coil erosion into the airway. Chronic steroid use (>30 days) is associated with delayed wound healing and an overall two to five times increase in wound complications.<sup>9</sup> When possible, endovascular therapy is generally avoided in patients with active vasculitis owing to the greater risk of complications.<sup>10</sup> For example, there



**Fig 3.** An image taken at the level of the distal trachea and right main stem bronchus with endoscopic scissors just before cutting the endovascular coil.

is a known increase in the complication rate in patients with systemic lupus erythematosus after endovascular or surgical interventions for intracranial aneurysms, likely



**Fig 4.** After discharge from the hospital, the patient was followed in clinic and radiographs were obtained approximately 1 month after right upper lobectomy. The posteroanterior radiograph demonstrates expected changed from right upper lobectomy, with unchanged appearance of the coil pack in the superior segment of the right lower lobe.

owing to active inflammation related to vasculitis and vessel wall fragility owing to chronic steroid use.<sup>11</sup>

## CONCLUSIONS

Although the initial coil embolization was life saving, this case illustrates a potential delayed complication of

endovascular treatment of inflammatory aneurysms, namely, migration of the coil through the inflamed walls into adjacent structures.

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