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Case Report

Bronchoscopy-guided removal of intrabronchial coil migration after coil embolization of pulmonary arteriovenous malformation☆,☆☆

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ABSTRACT

Pulmonary arteriovenous malformations develop in approximately 50% of hereditary hemorrhagic telangiectasia patients. Pulmonary arteriovenous malformations are often treated with coil embolization therapy. We report a case of a 45-year-old female with multiple pulmonary arteriovenous malformations due to underlying hereditary hemorrhagic telangiectasia who had undergone 14 coil embolization procedures over 16 years. She presented with sudden onset severe, unremitting, nonproductive cough from a foreign body sensation in the airway. Computed tomography of the chest demonstrated a metallic foreign body extending from the left lower lobe of the lung into the left mainstem bronchus and trachea. Bronchoscopy-guided removal of the foreign body revealed an intact embolization coil placed 8 years prior to presentation had partially migrated through the vessel and airway walls into the airway lumen, extending from the left lower lobe bronchus to the left mainstem bronchus. Coil migration is a rare, but potentially dangerous, complication of coil embolization therapy.

Introduction

Hereditary hemorrhagic telangiectasia (HHT) is characterized by mucocutaneous telangiectasias and visceral arteriovenous malformations (AVMs) in which arteries directly connect to veins without intervening capillaries [1]. HHT is inherited in an autosomal dominant fashion, and more than 80% of HHT cases result from mutations in the endoglin (ENG), activin A receptor type II-like 1 (ACVRL1), and SMAD4 genes [1,2]. The prevalence of HHT in the United States is approximately 12 per 100,000 [3]. HHT presents with a variety of symptoms depending on which organs are affected. The majority of HHT patients develop telangiectasias in the nasal mucosa, tongue, buccal mucosa, face, or hands, which are friable and prone to rupture with minimal stress [1]. Over 90% of HHT patients de-

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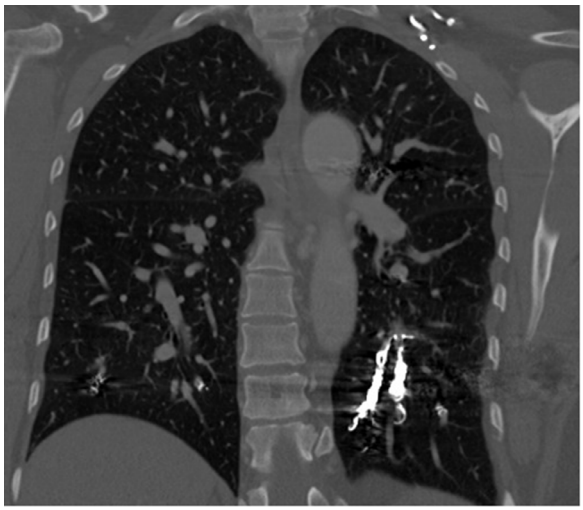

Fig. 1 – Coronal view, computed tomography (CT) of chest 9 months prior to presentation.
Chest CT showed satisfactory position of multiple embolization coils in both lungs.

velop recurrent epistaxis by the age of 21, which can range from mild, infrequent bleeding to gushing bleeds with minimal provocation [1,4]. Larger AVMs can also develop in the gastrointestinal tract, lungs, brain, and liver, which can lead to iron deficiency anemia, paradoxical embolization, hemorrhagic stroke, and high-output heart failure, respectively [1,5–8].

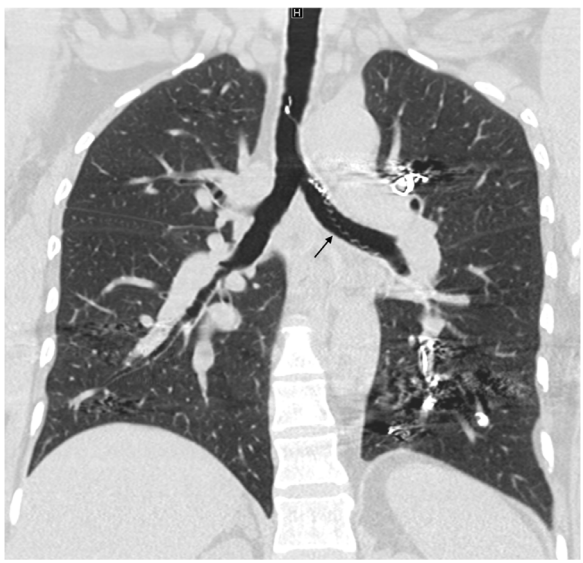

Fig. 2 – Coronal view, CT of chest on presentation.
Noncontrast chest CT on presentation demonstrated a metallic foreign body within the left mainstem bronchus (black arrow). This coil was used in a previous pulmonary AVM coil embolization in the left lung that subsequently migrated into the left mainstem bronchus.

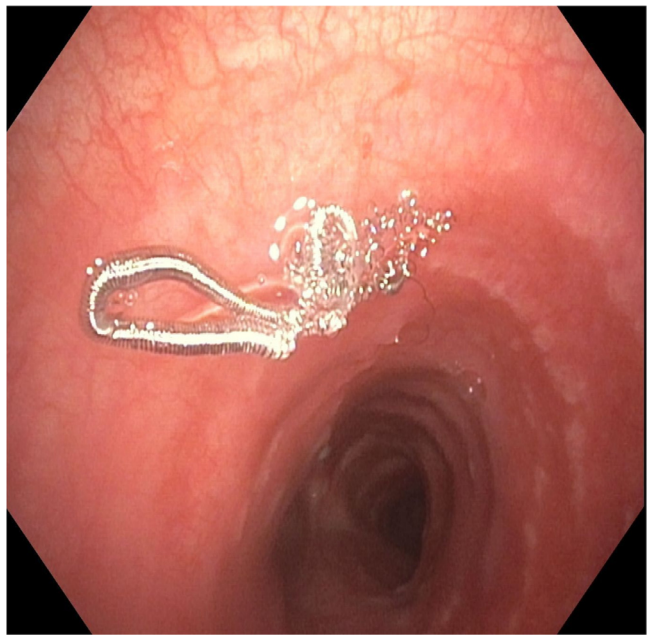

Fig. 3 – Bronchoscopy image.
Bronchoscopy within the trachea demonstrates the proximal extent of the metallic wire material. Direct mechanical clipping of coil was performed to remove the offending coil. After removal of the clipped coil, no other foreign bodies remained in the airway.

Pulmonary AVMs are one of the main sources of morbidity in HHT patients [7,9]. Approximately 50% of HHT patients develop pulmonary AVMs and complications arise in up to 70% of untreated pulmonary AVMs [7,9]. Complications of untreated pulmonary AVMs include ischemic stroke, brain abscess, dyspnea, hypoxemia, hemoptysis, and hemothorax [7,9]. In addition to the presence of the aforementioned complications, indications to treat pulmonary AVMs are a feeding artery diameter >2 mm or progressive increase in pulmonary AVM size [10]. Currently, the preferred treatment for focal, non-diffuse pulmonary AVMs is transcatheter embolization because of the ability to preserve pulmonary parenchyma and avoid major surgery and general anesthesia [9]. Metallic coils are often used to embolize pulmonary AVMs given their safety and maneuverability [9,11,12]. Coil embolization has been reported to have a 99% technical success rate, although known complications include coil recanalization or pulmonary AVM enlargement, which requires multiple percutaneous coil embolization attempts [11,12]. The most common complication of coil embolization of pulmonary AVMs is self-limited pleuritic chest pain [11,12]. However, other complications including vascular injury, cerebrovascular accidents, and coil migration have been reported [11,12]. At least 4 reports of intrabronchial coil migration have been published, 3 of which occurred 10 or more years after embolization [13–16].

Herein, we report a case of an HHT patient with pulmonary AVMs status post multiple embolization procedures with a complication of coil migration into the airway requiring bronchoscopic removal.

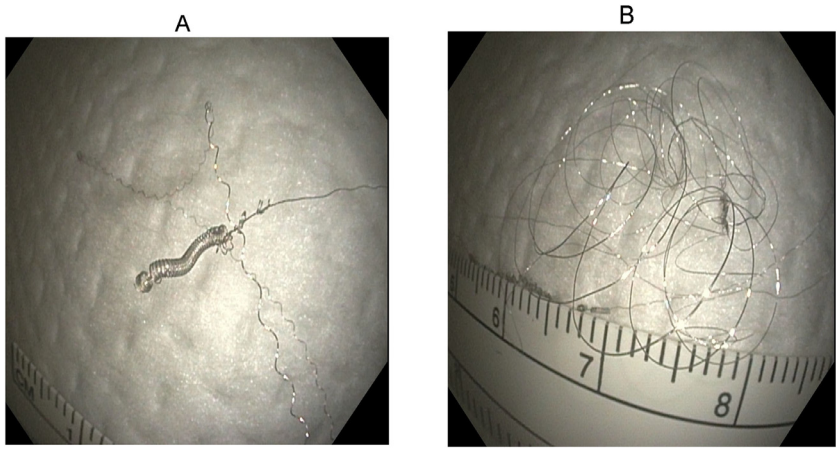

Fig. 4 – Retrieved coil material.
A. Retrieved coil fragment revealed partially unraveled coil material.
B. Retrieved coil fragment which had completely unraveled.

Case report

The patient was a 45-year-old female with clinically confirmed HHT based on recurrent epistaxis, first-degree family history, and multiple pulmonary AVMs. She was diagnosed at the age of 29 after having an embolic stroke and 2 transient ischemic attacks (TIA), which were caused by paradoxical embolization. Over the intervening 16 years, she underwent 14 coil embolization procedures for her pulmonary AVMs, the most recent being approximately 2 years prior to presentation. In total, 9 pulmonary AVMs in the right lung and 14 pulmonary AVMs in the left lung were embolized, some of which were embolized more than once due to recanalization. Chest computed tomography angiography (CTA) 9 months prior to presentation confirmed adequate and stable positioning of the coils (Fig. 1). However, on presentation, the patient presented with acute onset of unremitting, severe, nonproductive cough and a foreign body sensation in the airway. Chest computed tomography (CT) showed an elongated coil extending from the left lower lobe into the left mainstem bronchus and trachea (Fig. 2). This coil was originally placed at an outside hospital 8 years prior to presentation, thus the brand of the coil was unknown. The patient underwent same-day bronchoscopy, which readily identified unraveled coil material extending through the airway wall into the left lower lobe bronchus (Fig. 3A) as well as coil material embedded within the mucosa of the trachea (Fig. 3B). Using forceps, 3 pieces of coil were removed, ranging from 11.0 to 26.0 cm in length (Figs. 4A and B). Repeat chest CT confirmed no residual coil material in the airways, and that no pneumothorax occurred from the procedure (Fig. 5). Minimal bleeding occurred as a result of the mechanical removal of the coil. Hemostasis was confirmed after the procedure. The patient recovered uneventfully and the cough rapidly subsided; she was discharged home the next day and has not had recurrence.

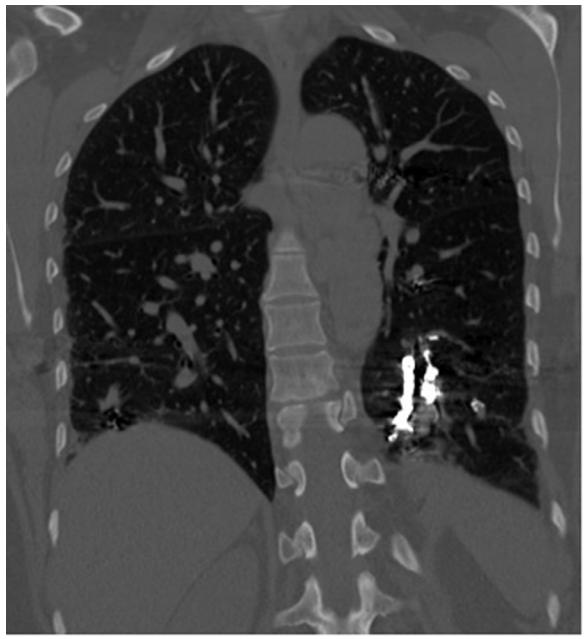

Fig. 5 – Coronal view, CT of chest status postcoil removal. Chest CT after bronchoscopy demonstrated no residual coil in the airway. No pneumothorax or bleeding occurred as a result of the bronchoscopy-aided removal of the migrated coil.

Discussion

This report describes a case of an HHT patient with multiple pulmonary AVMs treated by coil embolization who experienced coil migration into the airway requiring bronchoscopic removal.

Coil migration is a rare complication of coil embolotherapy [11–13]. Although few cases of coil migration from pul-

monary AVMs have been reported [13,14], coil migration after embolotherapy for other lung pathologies have also been documented. Cases include intrabronchial migration after coil embolization of pulmonary arterial aneurysms [17] and pseudoaneurysms [18] as well as from bronchial artery embolization [19]. Coil migration from pulmonary embolization can also cause more serious complications, such as fistulization [20], migration to the heart [11,21,22], pulmonary hypertension [11], pneumothorax [23], erosion into the bronchus [15], and migration into the contralateral pulmonary vasculature [22].

Various treatment options exist for managing coil migration, depending on the location of the migrated coil and the nature of the symptoms caused by the coil migration. If the patient remains asymptomatic or minimally symptomatic from coil migration and the coil is not threatening to erode into particularly sensitive tissues, some may opt to leave the migrated coil without any intervention [20,22]. If the coil migrates into the airway and is not fully expectorated, bronchoscopy-guided direct mechanical clipping may be utilized to remove the offending region of the coil [14,19]. Currently, there are no guidelines that exist for the management of the risk for bleeding secondary to coil migration. In the current literature, the decision to intervene on a possible bleeding risk is largely operator-dependent. In one case, to reduce the risk of bleeding from the communication between the pulmonary vasculature and the airway, the artery upstream of pulmonary AVM was embolized with histoacryl glue to completely cut off blood flow to the pulmonary AVM prior to the coil being mechanically removed [14]. In another case, minor bleeding was observed during bronchoscopic removal of the coil, but resolved spontaneously without requiring any hemostatic interventions [19]. If the migrated coil is difficult to reach, threatens to erode into sensitive structures, or poses a risk for massive bleeding upon coil removal, surgical resection has been employed [13,15,21–23]. In our case, since the coil migrated into the left main bronchus and trachea but could not be expectorated, the coil was mechanically removed using bronchoscopy.

Coils can migrate via several mechanisms. Some coils may migrate due to fluid mechanics as the coil stays intact and simply moves to a connecting vascular structure [21,22]. Coils may also erode through tissue parenchyma into adjacent structures as evidenced by coils migrating from vascular structures into the airway [13–15,19,20,23]. In our case, the coil eroded through the vessel wall and into the adjacent bronchus, as the chest CT showed the coil material extending from the original site of coil embolization in the left lower lobe to the left mainstem bronchus and trachea. Several studies have demonstrated that metallic devices implanted into internal organs or against mucosal surfaces can erode through soft tissue in both humans [24] and animal models [25,26]. In particular, many case reports have documented metallic coils eroding through various soft tissue structures, including the gastrointestinal tract [27,28], kidneys [29], esophagus [20], and pelvic structures [30].

Conclusion

In rare cases, coil embolotherapy in the pulmonary system can pose a risk for intrabronchial coil erosion and migration, which can occur years after the embolization procedure. For coils that migrate into the airway, bronchoscopy-guided mechanical removal of the coils can be utilized to extract the offending material while still preserving the lung parenchyma.

Patient consent

Written informed consent for the publication of this case report, including all imaging results, was obtained from the patient and is on file.

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