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Case report

Migrated coil expectorated 12 years after embolization of pulmonary arteriovenous malformation, due probably to abscess formation around the coil

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ABSTRACT

Pulmonary arteriovenous malformations (PAVMs) are rare vascular structures providing direct capillary-free communications between pulmonary arteries and veins. Embolotherapy is indicated as a front-line therapy. We report an unusual long-term complication of coil embolization for a 44-year-old woman with hereditary hemorrhagic telangiectasia (HHT) who had repeatedly undergone the procedures for her PAVMs. She expectorated the coil which had been placed 12 years earlier and migrated to the bronchus according to the chest radiogram and bronchoscopy. Histology of the resected lung segment suggested the cavity communicating with the bronchus was the consequence of abscess formation around the coils. Even after technically successful embolization to PAVMs, long term follow-up should be necessary paying attention to the symptoms and imaging to avoid massive hemoptysis and subsequent emergency surgery.

1. Introduction

Pulmonary arteriovenous malformations (PAVMs), providing direct capillary-free communications between pulmonary arteries and veins, cause easy access of septic or non-septic emboli to systemic circulation and right-to-left shunt of unoxygenated blood, in addition to hemoptysis or hemothorax by PAVMs rupture [1]. By considering the patient factors such as the feeding artery diameter, PAVM-related symptoms, and the patient's ability to tolerate the procedure, timing and methods of intervention should be determined [2]. Embolotherapy demonstrated better outcomes rather than surgery in terms of success rate and complications [3]. Among other materials, coils have been used for embolization to PAVMs although long-term complications related to coil embolization have been reported such as endobronchial coil migration [4,5], hemoptysis [6], or worsening of pulmonary hypertension [7].

We report a woman expectorating a coil from the cavity, communicated with the bronchi 12 years after embolization of PAVMs.

2. Case report

A 44-year-old Japanese woman presented with strong dry cough producing a thin wire (Fig. 1). Her past medical history was significant for hereditary hemorrhagic telangiectasia (HHT). Twelve years earlier, she underwent first coil embolization of multiple PAVMs in the right S^9 (Fig. 2) after massive hemoptysis. The first procedure was technically successful and confirmed no coil migration on the next day (Fig. 3A). To treat recanalization of PAVMs, confirmed by angiography, and enlargement of untreated PAVMs, second coil embolization was performed ten years after the first time in the right S^9. Since she was diagnosed with asymptomatic stroke by MRI during the follow up, a possible paradoxical thrombus, the third coil embolization was perfomed for untreated AVM located left S6 six months before coil expectoration. The chest X-ray, one day before the third coil embolization, demonstrated coil deformation (Fig. 3B). One the day she expectorated the thin wire, chest images confirmed one of the packed coils coming out in the form of a loop toward the proximal side (Fig. 3C). Bronchoscopy

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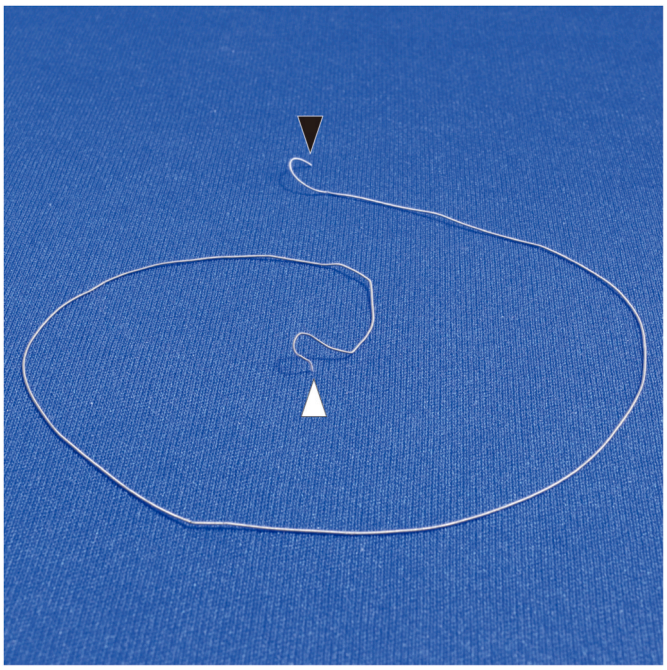

Fig. 1. The thin wire
The detachable coil in length 25 cm (DETACH DCS Coil, Cook Japan, Tokyo, Japan). Originally, distal end of the coil shaped J. There is no deformation, disconnection, or un-raveling. White arrowhead: proximal end of coil. Black arrowhead: distal end of coil.

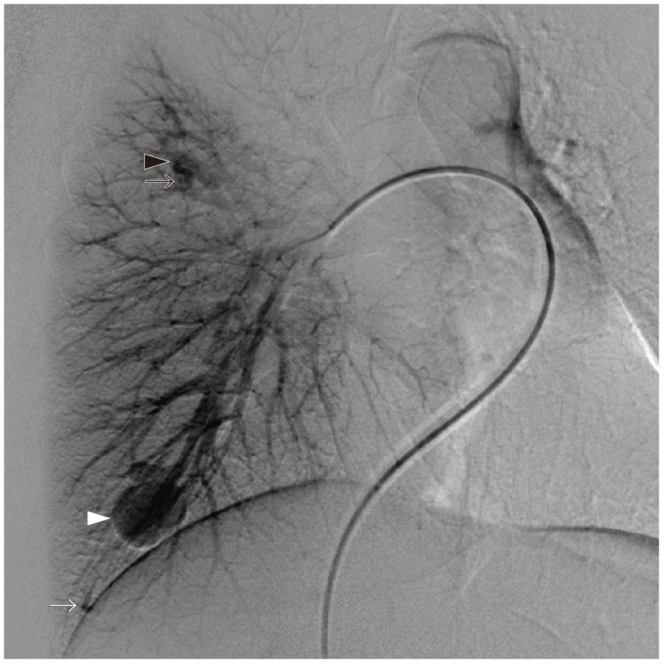

Fig. 2. Angiography of pulmonary arteriovenous malformations
Pre-embolization angiography of the right pulmonary artery. Four pulmonary arteriovenous malformations (PAVMs) was detected in two segments, right S^9 (white arrow and white arrowhead) and right S^6 (black arrow and black arrowhead). As for the largest PAVM in right S^9 with a one feeding artery of 4.6 mm and nidus measuring 26.2 mm (maximum diameter).

confirmed another thin wire located in the right B^9 bronchial lumen (Fig. 4). We had not done any microbiological tests at that time. The thin wire was thought to be the coil used for embolization 12 years before. Since we were afraid of communication between the respiratory tract and blood vessels, and thoracoscopic right basal segmentectomy ($S^{7\text{-}10}$) was performed instead of pulling out of wire. Histology revealed that the cavity, containing debris with chronic inflammation and opening to the right B^8 and B^9, exhibited no communication with the vasculature, such as the right A^{8a} or V^{9b}, which ended beside the cavity (Fig. 5).

3. Discussion

As for embolotherapy, metallic coils are most commonly used because they are easy to control and operable to occlude both small and large arteries [8,9]. The technical success rate of coil embolization for PAVMs is extremely high (≥99%) and coil embolization-related complication is rare (≤0.65%) [10,11]. However, less is known about the long term complications, necessitating careful observation of coil migration for many years like this presented case. The coil migration to bronchi may cause two types of troubles: a bronchial pulmonary artery fistula and a bronchial foreign body. Since the coil was placed in the vessel, fistula formation by coil migration may induce fatal massive hemoptysis [12], air embolism [13] or retrograde infection to the blood stream. It has also been reported that the metallic coil as a foreign body induced cough and hoarseness [14].

Microscopic examination of the resected specimen revealed the coils were located in the cavity containing debris with chronic inflammation (Fig. 5). Histology demonstrated the cavity had fibrotic wall opening to the bronchi (rt. B^8 and B^9) with no communication to the interrupted embolized vessels (rt. A^{8a} and V^{9b}). We hypothesized that the coil had protruded from PAVM nidus and injured lung parenchyma, causing infection. The coils prevented healing of the infection and sustained chronic inflammation. In the process of tissue repair, the embolized vessels were occluded by fibrosis.

Ball et al. hypothesized that relatively large volume change of the lower lung by breathing could help coil migration [15]. In our case, the PAVM nidus was located in the S9, the lower lung, and affected by mechanical pressure or shear stress intermittently with breathing, leading to the rupture of the weakened wall and coil migration.

To monitor recanalization after coil embolization, the guidelines stated 'ultidetector thoracic CT with thin-section reconstruction (1–2 mm) should be undertaken within 6–12 months after embolization and then approximately every 3 years' [2], or MRA instead of CT to reduce coil artifact. As for coil migration, chest X-ray image is most suitable modality for evaluation of coil shape and position. More than ten years follow-up would be needed. At the same time, we need to inform patient about coil migration which could occur as a late complication.

When coil migration was suspected, the following must be checked; 1) medical interview about cough, sputum; 2) vital signs and physical examination including auscultation; 3) enhanced CT/magnetic resonance angiography (MRA), or angiography if needed; 4) bronchoscopic confirmation and bacterial culture test s. However, coil artifact in CT makes it difficult to figure out precise hemodynamic status in clinical practice [16]. In our case, cavity-artery communication was not detected either in the imaging of pre-surgical screening or post-surgical histopathological examination.

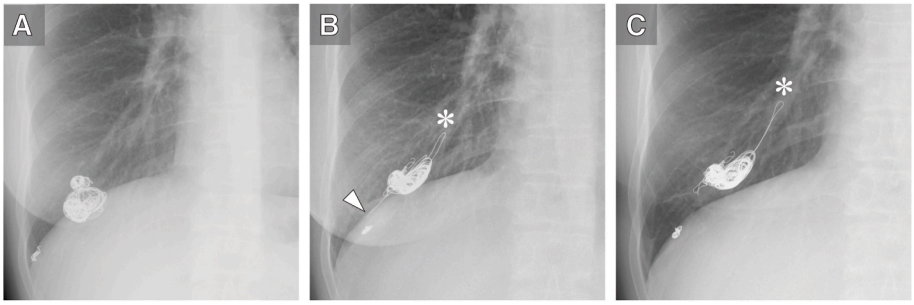

Fig. 3. Chest radiography
A) A day after first coil embolization. Two pulmonary arteriovenous malformations (PAVMs) in the right S^9 were filled up with the total of sixteen detachable coils (DETACH DCS Coil or Cook Embolization Coil Tornado, Cook Japan, Tokyo, Japan).
B) A day before third coil embolization. There were coil compaction and deformation. One coil extended forward to proximal bronchi (*). The other coil extended forward to distal bronchi (white arrowhead). C) On the day she producing a thin wire. There remains the coil extended forward to central bronchus in the right B^9 (*).

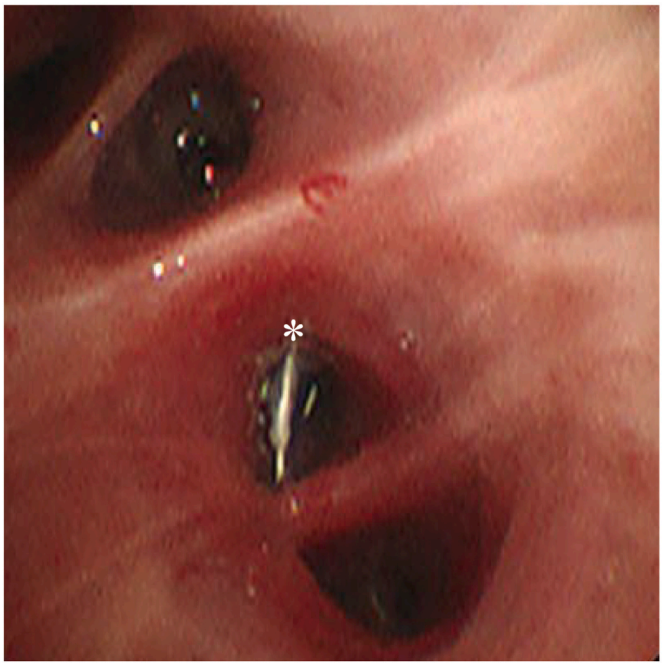

Fig. 4. Bronchoscopic findings
The thin looped wire was seen at right B^9 (*). There were redness and swelling of the right B^8 and B^9 bronchial wall. No hemoptysis or purulent sputum were seen.The wire moved back and forth with the breath.

Treatment algorithm for coil migration have not been established. Based on the examination, therapeutic strategy should be decided case by case considering patients' requests. There is a previous report on bronchoscopic removal of migrated coil after bronchial artery embolization. The authors carefully split the coil by a loop cutter and removed by forceps [14]. We did not choose transbronchial coil removal under intubation because of the possibility of massive hemoptysis.

For the treatment of hemoptysis by coil migration, the confirmation of the feeding artery, pulmonary or bronchial, is necessary before transcatheter embolization. To our knowledge, there have been three case reports about coil migration after embolotherapy for PAVMs. Lobectomy was performed in two cases after repeated bronchial artery embolization failed to control hemoptysis [5,6]. Lobectomy was also performed in the other case without any symptoms although the authors reconsidered their hasty decisions [4].

More than two-thirds of HHT patients were diagnosed before the age of 40 years [17], with multiple PAVMs possibly occurring afterwards [18]. Maintaining respiratory function is of vital importance in relatively young patients with HHT. We, therefore, think that indication of non-emergency surgery for patients with minimal symptoms should be decided with careful consideration. Since our patient had suffered massive hemoptysis years before, we suspended bronchoscopic coil removal to avoid life-threatening hemoptysis, and selected basal segmentectomy to minimize the risk and an area of resection.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

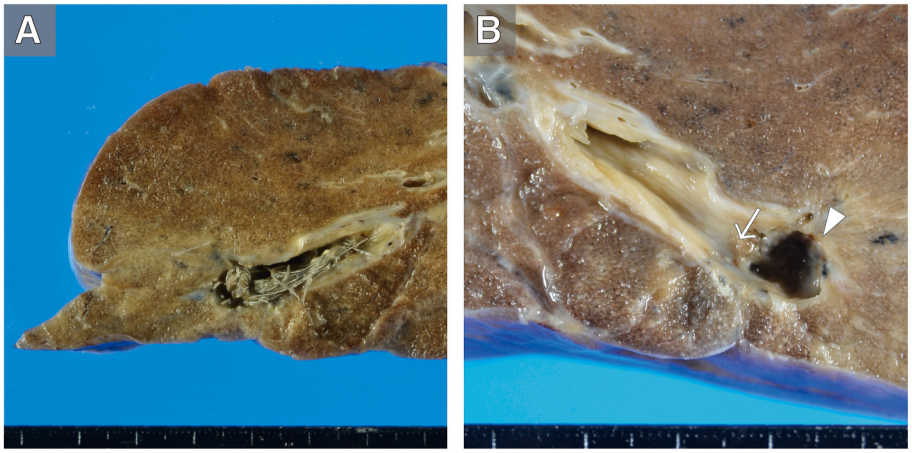

Fig. 5. Photograph of the resected right lower lobe
A) Coils in the cavity and bronchiectasis of the right B^{9b}, which were covered with the debris. B) A cavity lesion (arrowhead) was perforated (arrow), and in communication with the bronchus. Histology demonstrated the cavity had fibrotic wall opening to the bronchi (rt. B^8 and B^9) with no communication to the interrupted embolized blood vessels (rt. A^{8a} and V^{9b}).

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