

Case Report

Two cases of cryptogenic life-threatening hemoptysis – identification and management of bleeding point

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Cases: Case 1: A 63-year-old woman was referred for coughing blood. Although cardiorespiratory dynamics were stabilized by artificial respiration under sedation, severely poor ventilation developed from asphyxia associated with massive respiratory tract hemorrhage. One-lung ventilation was temporarily secured by endotracheal tube insertion into the left main bronchus just prior to cardiopulmonary arrest.

Case 2: A 72-year-old man was referred for massive hemoptysis after coughing, then intubated and placed on a respirator. During angiography, blood clots collected with bronchoscopy confirmed extravascular leakage into the right main bronchus.

Outcomes: Both showed no hemoptysis recurrence after bronchial artery embolization and were discharged. Case 1 required intensive treatment for 6 days, including artificial respiratory management.

Conclusion: Emergency one-lung ventilation was required for asphyxia in Case 1, and we had difficulties with bleeding point identification and hemostatic therapy. From that experience, we noted hemoptysis during angiography using bronchoscopy in Case 2, enabling prompt bronchial artery embolization.

Key words: bronchial artery embolization, bronchoscopy, extravasation, life-threatening, massive hemoptysis

INTRODUCTION

WE REPORT TWO cases with good outcomes following successful ventilation maintenance during asphyxia and airway management following bleeding point identification, combined with hemostatic therapy using bronchial artery embolization (BAE). Similar cases are reviewed.

CASES

Case 1

A HEALTHY NON-SMOKING 63-year-old woman was examined for coughing blood. Detailed findings

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including upper gastrointestinal endoscopy denied hematemesis. Respiratory status worsened with hemoptysis and she was referred to us under artificial respiration and endotracheal intubation. Chest X-ray and computed tomography findings at admission revealed a right lung field infiltrative shadow (Fig. 1A). Bronchial angiography showed dilatation >2 mm on the proximal side of the right main bronchial artery. Diffuse angiectasis >0.5 mm was also depicted on the distal side (Fig. 1B) and bronchoscopic findings showed massive blood clots in the right main bronchus (Fig. 1C). However, hemoptysis cause, such as tumors, could not be determined. The patient was admitted to the intensive care unit (ICU) for observation; hemostasis was confirmed by bronchoscopy, although the bleeding amount was unknown, while cardiorespiratory dynamics under sedation were stable. The next day, she suffered massive hemoptysis triggered by coughing and showed severely poor ventilation caused by asphyxia. One-lung ventilation was obtained by inserting an endotracheal tube into the left main bronchus to temporarily secure ventilation (Fig. 1D). Following emergency bronchial angiography, the culprit vessel was detected using a microcatheter. Bronchial artery

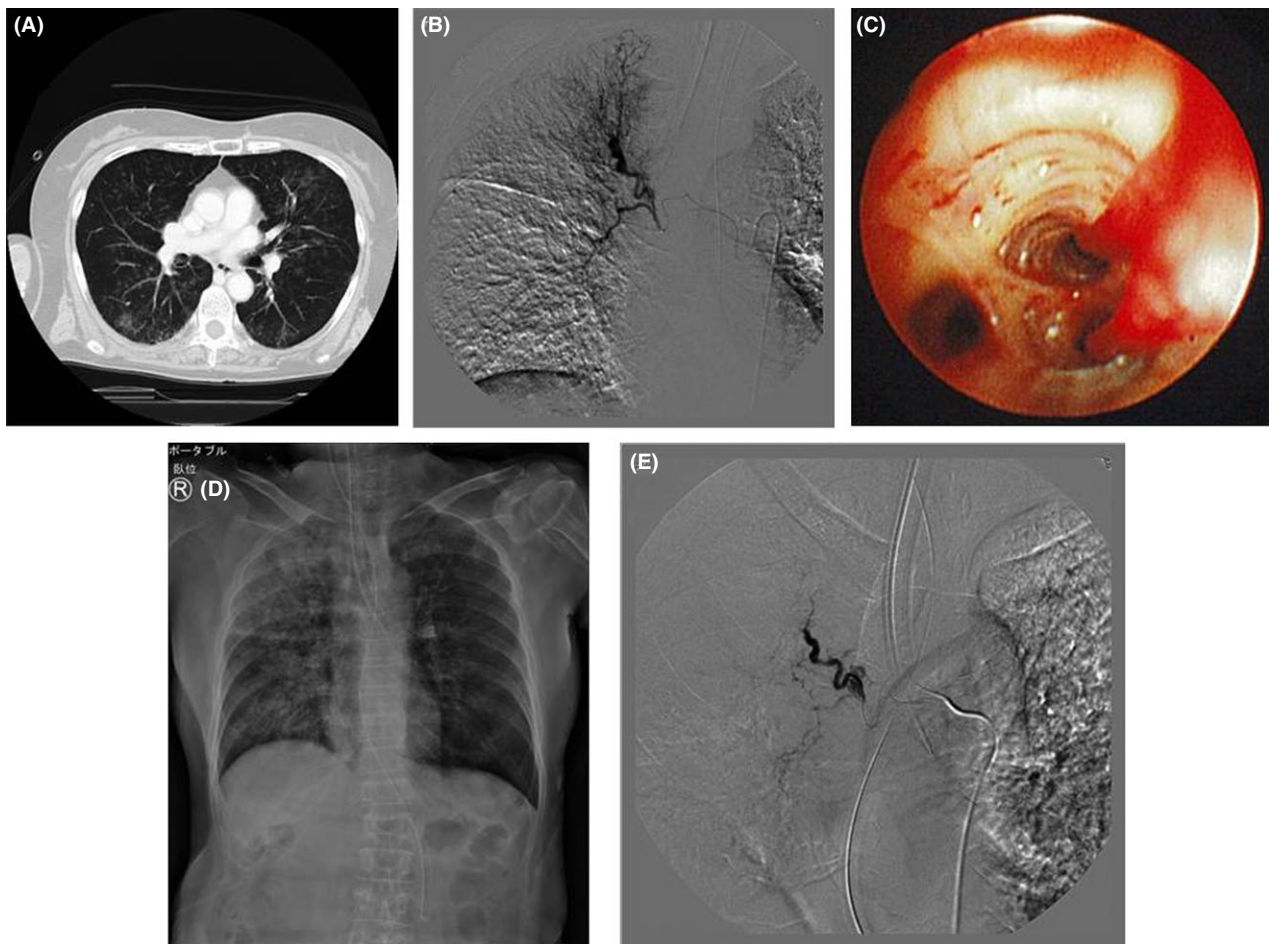


Fig. 1. A, Computed tomography findings in a 63-year-old woman at admission revealed an infiltrative shadow in the right lung field. B, Bronchial angiography showed dilatation >2 mm on the proximal side of the right main bronchial artery and diffuse angiectasis >0.5 mm on the distal side. C, Bronchoscopic findings showed massive blood clots in the right main bronchus. D, The day after admission, the patient suffered massive hemoptysis triggered by coughing and severely poor ventilation caused by asphyxia. One-lung ventilation was obtained by temporarily inserting an endotracheal tube into the left main bronchus to secure ventilation. E, Following emergency bronchial angiography, the culprit vessel was detected by use of a microcatheter. Bronchial artery embolization was carried out.

embolization was carried out using gelatine sponge particles (Fig. 1E) and the endotracheal tube was repositioned to ~ 4 cm from the trachea bifurcation. The patient was then returned to the ICU. As anemia progressed, a transfusion was carried out on hospital day 4, after which she was weaned from ventilatory support on day 6 and discharged on day 13.

Case 2

A 72-year-old man with a history of diabetes and smoking vomited up blood a few hours after the subjective sign of coughing. He was transported on an emergency basis to our

center as a referral. Respiratory status was stable on admission and he underwent chest contrast-enhanced computed tomography without intubation, which revealed only ground-glass opacity dominant in the right lung field (Fig. 2A). No vascular abnormality nor extravascular leakage was noted. An episode of massive hemoptysis (200 mL in 4 h) was confirmed, thus BAE was prepared while carrying out endotracheal intubation and providing artificial respiratory care for enabling bronchoscopy during interventional radiology. As in Case 1, vascular dilatation >2 mm was found on the proximal side, while the vascular abnormality in the periphery showed diffuse spreading (Fig. 2B). The hemoptysis-causing vessel and bleeding point could not be

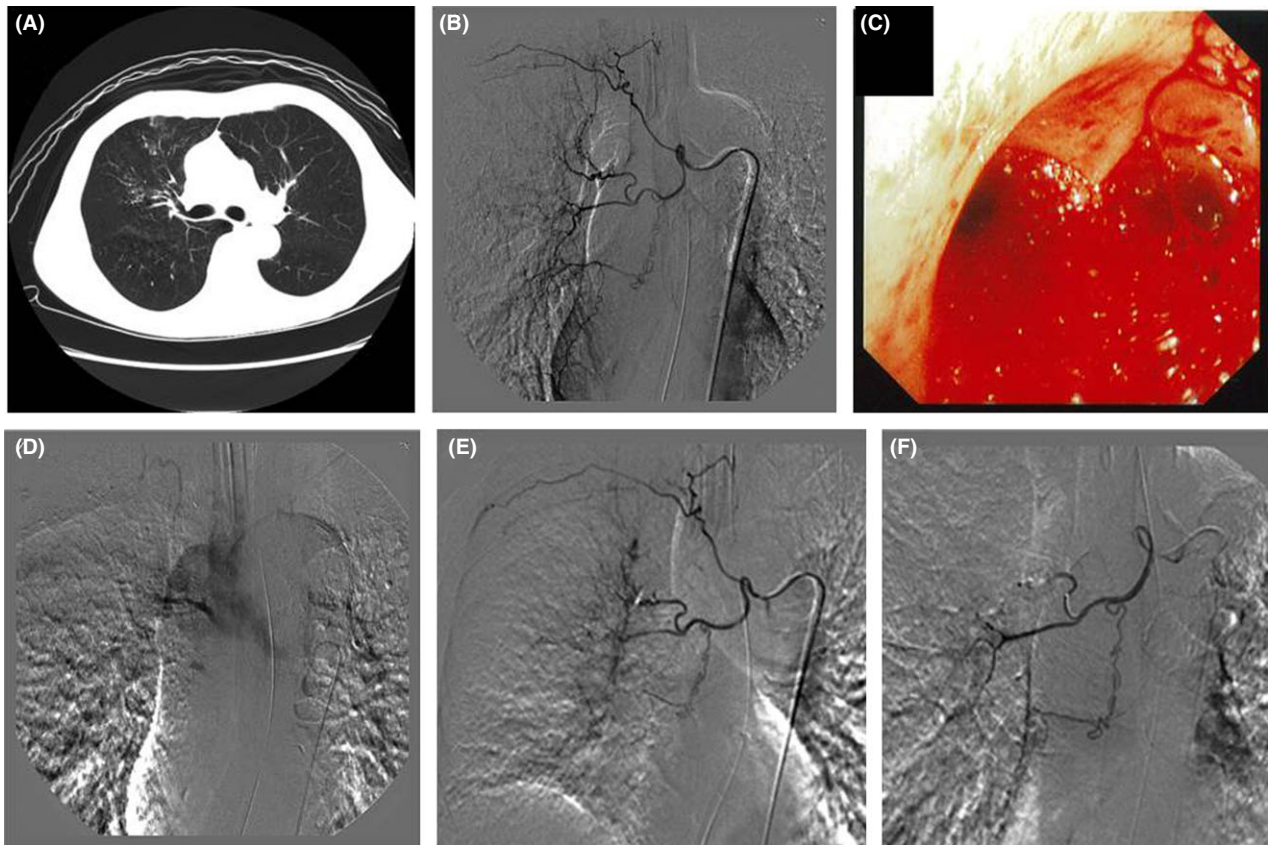


Fig. 2. A, Contrast-enhanced computed tomography findings of a 72-year-old man revealed only ground-glass opacity that was dominant in the right lung field. No vascular abnormality nor extravascular leakage was noted. B, Vascular dilatation >2 mm was found on the central side; the vascular abnormality in the periphery showed diffuse spreading. C, D, While securing the safety of the airway, blood clots were aspirated with a bronchoscope during angiography, which resulted in confirmation of massive foamy hemoptysis. Intercostal angiography at that time also revealed extravascular leakage from the lobar bronchus to right main bronchus. E, Angiographic findings after airway suctioning showed visible changes in the vascular abnormality in the periphery of the right bronchial artery. Thus, the culprit vessel causing massive hemoptysis was identified by removing blood clots. F, Embolization of the right bronchial artery was carried out.

located for undertaking BAE. While securing airway safety, blood clots were aspirated with bronchoscopy during angiography, resulting in confirmation of massive foamy hemoptysis (Fig. 2C). Intercostal angiography carried out at that time also revealed extravascular leakage from the lobar bronchus to right main bronchus (Fig. 2D, Video S1). Because of anastomosis of the intercostal artery and the right bronchial artery as the causative vessel, we considered that contrast media might leak from the lesion into the airway. A comparison of angiographic findings of the right bronchial artery before and after aspiration confirmed visible changes in the vascular abnormality (Fig. 2B, E) in the periphery after removing blood clots, and the culprit vessel causing massive hemoptysis was identified. Embolization of the right bronchial artery was carried out using Gelfoam

(Fig. 2F). Immediately after interventional radiology completion, the endotracheal tube was removed. The patient returned to the ICU for follow-up observations and no hemoptysis recurrence was noted. Bloody expectoration improved and he was discharged on hospital day 10.

DISCUSSION

IT HAS BEEN reported that hemoptysis from the bronchial arterial system accounts for 90% of airway hemoptysis cases,^{1,2} for which BAE has been established as a therapeutic option. However, the utility of bleeding site identification by bronchoscopy before BAE may be limited and the procedure might not always be necessary,³ although extravascular leakage detection provides convincing

evidence. In Case 2, BAE was carried out, as we detected extravascular leakage during blot clot removal by bronchoscopy while monitoring changes in angiography of the culprit vessel in the bronchial artery periphery (Fig. 2B, E).

Bronchial arterial dilation ≥ 2 mm on the proximal side and ≥ 0.5 mm on the distal side indicate a vascular abnormality,⁴ with tortuosity and hyperplasia also generally seen. Bronchial artery embolization is generally accepted for such evidently abnormal vessels. However, paraplegia resulting from embolization of the artery of Adamkiewicz is widely known as a complication of BAE, although the incidence is as low as 1%.^{2,5} This causes hesitation in proceeding with BAE if there is doubt that it is necessary. As seen in Case 1, although a vascular abnormality was identified in the initial angiography findings, it was difficult to determine BAE because of a lack of information regarding bleeding amount.

The frequency of extravascular leakage detection by bronchial arteriography for providing evidence for BAE is not high regardless of the clinical situation,^{6,7} as that has been reported in 5 of 140 (3.6%)⁸ and 3 of 28 (10.7%)³ cases. Moreover, no known reports have confirmed extravascular leakage during a bronchoscopic examination for removing blood clots. Accordingly, it is significant that we clarified extravascular leakage using the present method. In Case 1, comparisons of angiograms performed twice failed to confirm changes in the culprit vessel in the right bronchial artery periphery (Fig. 1B), which also occurred in Case 2 (Fig. 2B, E). As hemostasis with one-lung ventilation was effective in the second angiography procedure in Case 1, it is likely that changes shown in Case 2 revealed minute extravascular leakage.

Asphyxia is a physiological mechanism showing worsening of airway hemoptysis, with massive airway hemoptysis defined as 100 mL/24 h or more.⁹ In 2008, Ibrahim proposed that this condition should be recognized and termed not as “massive”, but rather “life-threatening” hemoptysis.¹⁰ In Case 1, bronchial angiography was undertaken twice before deciding on BAE. During our decision-making process, the patient experienced potentially life-threatening worsening due to asphyxia. However, in Case 2, we soon recognized the life-threatening state of airway hemoptysis, even though it was massive hemoptysis, and promptly carried out bronchoscope-assisted BAE under general anesthesia, which also shortened the periods of artificial respiration and hospitalization.

Our procedure has limitations, including risk of asphyxia, although that is highly predictable when aspirating with a bronchoscope. Accordingly, countermeasures such as control of bleeding from the airway side by using a blocker and

quick introduction of extracorporeal circulation with an oxygenator should be prepared.

CONCLUSION

WE TREATED MASSIVE airway hemoptysis by extensive airway care including one-lung ventilation, and observation and treatment with a bronchoscope. From our experience with difficulty in identifying the bleeding point and performing hemostatic therapy in Case 1, we promptly commenced therapy with BAE in Case 2, based on identification of bleeding with bronchoscopy during angiography.

CONFLICTS OF INTEREST

NONE.

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SUPPORTING INFORMATION

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

Video S1. Case 2 of life-threatening hemoptysis in a 72-year-old man. While securing the safety of the airway, blood clots were aspirated with a bronchoscope during angiography, resulting in confirmation of massive foamy hemoptysis.