

# Massive Hemoptysis : Be Aware of Non-Bronchial Systemic Arterial Supply – A Case Report and Literature Review

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Massive hemoptysis is a life-threatening emergency. The bleeding usually originates from bronchial arteries (BAs), but may also arise from pulmonary arteries, or less frequently from non-bronchial systemic arteries (NBSAs). Bronchial artery embolization (BAE) is considered the first-line management for hemostasis; however, the high rebleeding rate remains a challenge. Bleeding from NBSAs is frequently missed, and this can lead to treatment failure. Surgical resection of the affected lung may eventually be required. We reported the case of a patient with tuberculous lung destruction in the right upper lobe (RUL), who experienced massive hemoptysis from both the BAs and an NBSA. Despite successful embolization of the BAs, bleeding recurred and led to cardiac arrest 2 days after the BAE. With the support of extracorporeal membrane oxygenation, the patient underwent an emergency lobectomy of the RUL. However, re-operative surgery was subsequently required due to rebleeding from a NBSA arising from the right subclavian artery, which was initially missed. The patient survived the critical bleeding episodes, but eventually died of recurrent pneumonia. Comprehensive imaging examinations, recognizing the imaging features and risks of recurrent bleeding, and planning appropriate treatment modalities are essential to improving the outcome in cases of massive hemoptysis. (*Thorac Med* 2018; 33: 130-138)

Key words: bronchial arterial embolization, massive hemoptysis, multidetector computed tomography, non-bronchial systemic artery

## Introduction

Massive hemoptysis is uncommon, but can be life-threatening. Uncontrolled bleeding is associated with a mortality rate of more than 50% [1-2]. Essential management consists of stabili-

zation, securing the airway, and identifying the bleeding site and cause, followed by effective hemostasis. Massive hemoptysis has been characterized as varying from 100 to 1,000 mL of expectorated blood within a 24-hour period [3-5] or more than 150 mL in 1 hour [5]. In 90%

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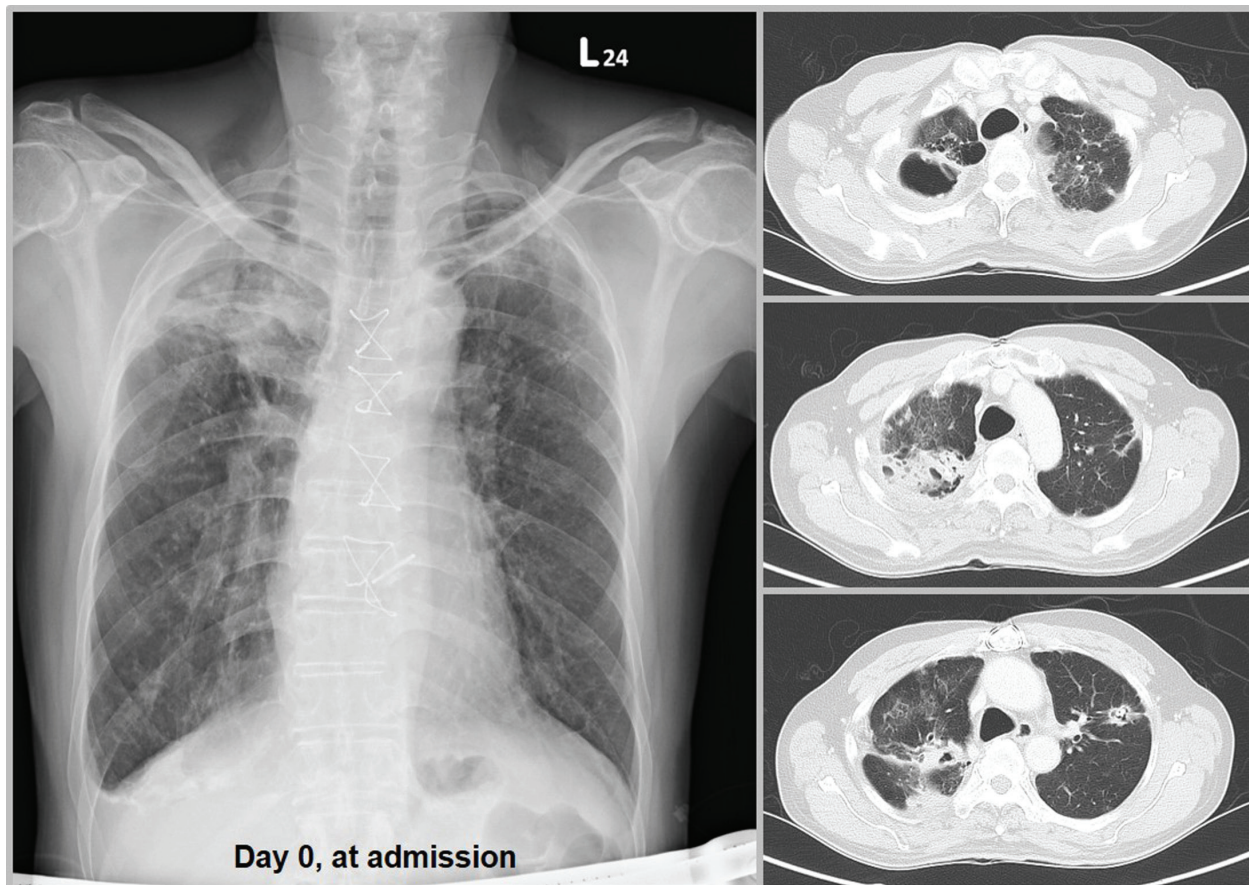
of cases, bleeding originates from the bronchial arteries (BAs). However, it may occasionally originate from pulmonary arteries (5%) or non-bronchial systemic arteries (NBSAs) (5%). Multidetector computed tomography (MDCT) has emerged as an essential tool to identify the bleeding site and possible cause, with the option of vascular mapping if MDCT angiography (MDCTA) is available [6-8]. Bronchial artery embolization (BAE) is currently considered to be the first-line intervention, with an immediate control rate of more than 80%, depending on the underlying disease [7-8]. However, early recurrence may be noted in up to 10-33% of cases, especially when ectopic branches of BAs, NBSAs, and pulmonary arteries are involved [7,9]. Although surgery is the final resort to control bleeding, emergency surgery for hemostasis is associated with high mortality (25-40%) [4-5,10-11]. Successful management of massive hemoptysis relies on expeditious diagnosis and appropriate intervention, which remains a challenge for clinicians. We report a patient with massive hemoptysis, who underwent a seemingly successful BAE, but early recurrence occurred due to a missed NBSA. The imaging pitfalls and need to choose the right management modalities are discussed in this report.

## Case Report

A 63-year-old male, who was a heavy smoker, was admitted with complaints of mildly persistent hemoptysis for 1 week. He was an alcoholic and intravenous drug abuser. Pulmonary tuberculosis (PTB)-related lung destruction in the right upper lobe (RUL) and cylindrical bronchiectasis of both lungs were diagnosed previously (at the age of 58 years), when he was scheduled to undergo aortic valve

replacement and aortic root graft implantation. Following the surgery, he was under long-term anticoagulation treatment with warfarin.

In this hospitalization, the chest radiograph (CXR) and MDCT at admission revealed typical changes to the old PTB with superimposed new consolidation in the RUL and superior segment of the right lower lobe (Figure 1). Blood test results were unremarkable except for coagulopathy. Warfarin was discontinued. Coagulopathy was corrected after a supplementary vitamin K injection and fresh plasma transfusion. His symptoms improved with conservative treatment. However, massive hemoptysis abruptly occurred on day 9. He was intubated and underwent an emergency BAE. The CXR showed an enlarged patchy infiltration in the RUL (Figure 2A). Thoracic aortic angiography revealed hypervascularity in the RUL with dual blood supply, mostly from the BAs and partly from the NBSA; this was not considered to be of clinical significance and was thus ignored (Figure 2B and 2C). After super-selection and embolization with gelfoam cubes for the BAs, the hypervascular area was markedly reduced (Figure 3) and hemoptysis subsided. The patient was scheduled to undergo RUL lobectomy, but it was postponed due to the unavailability of a surgical facility. Massive hemoptysis recurred, unfortunately, leading to cardiac arrest on day 11. With successful resuscitation, venovenous extracorporeal membrane oxygenation (VV-ECMO) was set up. A rescue fibrobronchoscopy failed to maintain airway patency because of sustained hemoptysis. A double-lumen endotracheal tube insertion and an emergency RUL lobectomy were carried out. Surgical findings revealed engorged BAs and active extravasation in the RUL. Post-operative fibrobronchoscopy revealed no further endobronchial bleeding.



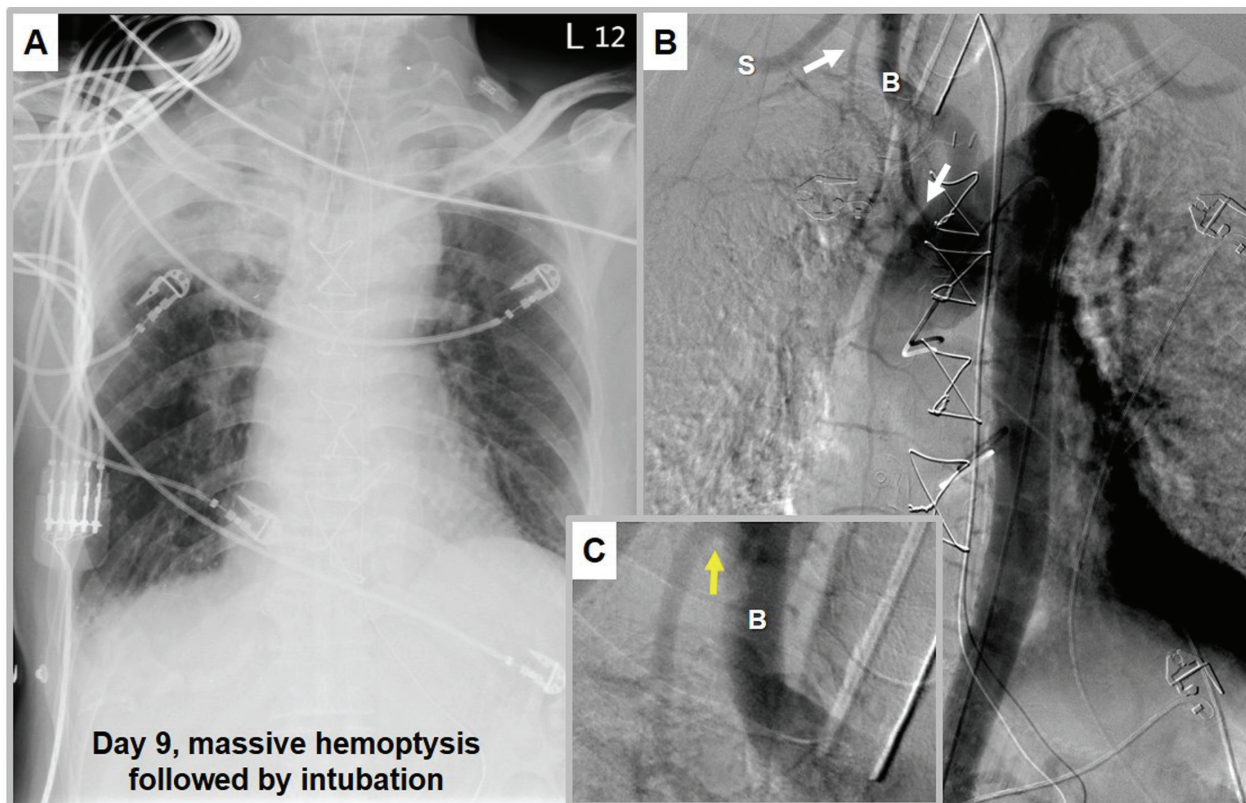
**Fig. 1.** Thoracic images at admission. Plain film revealed consolidation, cavitation, and volume reduction in the right upper lobe; bilateral apical pleural thickening; right costophrenic blunting; and bronchiectasis of both lungs (left panel). Multidetector computed tomography (lung windows) highlighted the significant pleural thickening and bronchiectasis with considerable secretion retention in the bronchiectatic area (right panel).

However, 5 hours later, excessive bleeding from the chest tubes was noted, and emergency exploratory thoracotomy was performed to evaluate the cause of bleeding. An NBSA arising from the base of the right subclavian artery in the pleural thickening of the right apex was found to be the cause of bleeding. Hemostasis was achieved after ligation of this NBSA. On re-evaluation of the contrast MDCT, the NBSA and hypervascular vessels in the thickening pleura – which were initially not recognized – could be identified (Figure 4). Subsequent pathological examination confirmed the diagnosis of bronchiectasis with acute and chronic pa-

renchymal hemorrhage, and pleural/subpleural fibroelastosis with acute suppurative pleuritis. Examinations for fungal and tuberculosis infection revealed negative results. He survived the critical bleeding episodes, and VV-ECMO was successfully discontinued on day 14. However, he died of recurrent ventilator-associated pneumonia on day 25.

## Discussion

We presented the case of a patient with PTB-related lung destruction, complicated with recurrent massive hemoptysis due to an

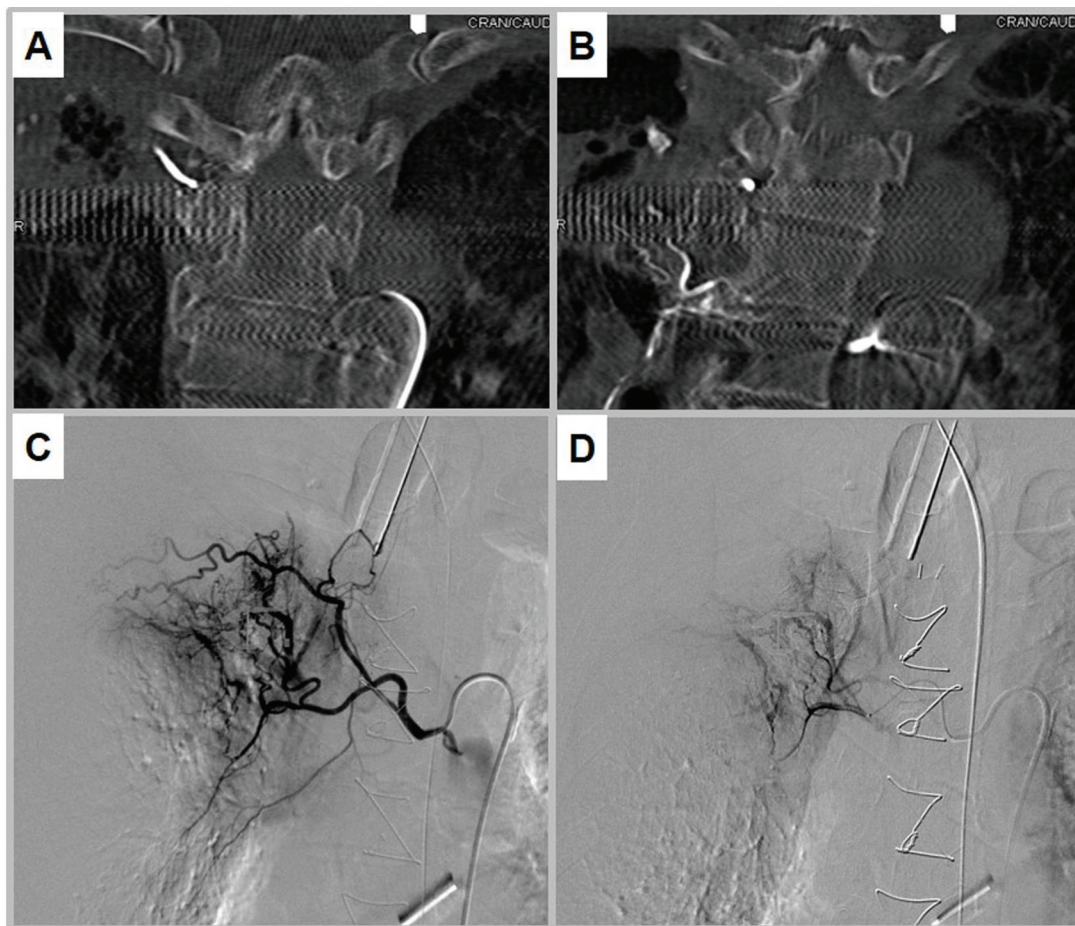


**Fig. 2.** Thoracic images after massive hemoptysis. A. The plain film showed a new patchy infiltration in the right upper lobe. B. The emergency thoracic aortography showed hypervascularity of the right upper lobe, with dual blood supply from a non-bronchial systemic artery (upper white arrow) and bronchial arteries (lower white arrow). C. The higher magnification image illustrates the non-bronchial systemic artery (yellow arrow) originating from near the bifurcation of the brachiocephalic (B) and subclavian artery (S).

uncommon condition – dual blood supply from the BAs and the NBSA. Failure to identify and manage the NBSA potentially contributed to early recurrence of massive hemoptysis and subsequent hypovolemic shock.

In addition to technical failure, early recurrence after BAE often results from incomplete identification of potential bleeding sources, which can include pulmonary arteries, ectopic or collateral branches of BAs, and an NBSA from the thoracic or abdominal artery near the affected lung [7,9]. Massive hemoptysis from an NBSA should be considered in patients with a history of chronic lung disease with pleural

involvement. Two CT signs suggestive of the presence of NBSAs are pleural thickening (>3 mm) adjacent to the parenchymal lesion, and enhancing vascular structures within the extrapleural fat layer, which have a sensitivity and specificity of 80% and 84%, respectively [12]. The origins of an NBSA usually depend on the location of the affected lung segments, such as the subclavian artery in the apices, the intercostal artery in the posterior segments, the internal thoracic artery in the anterosuperior segments, and the inferior diaphragmatic artery in the bases [7,13]. In this case, regrettably, we neglected to identify the suggestive signs in the

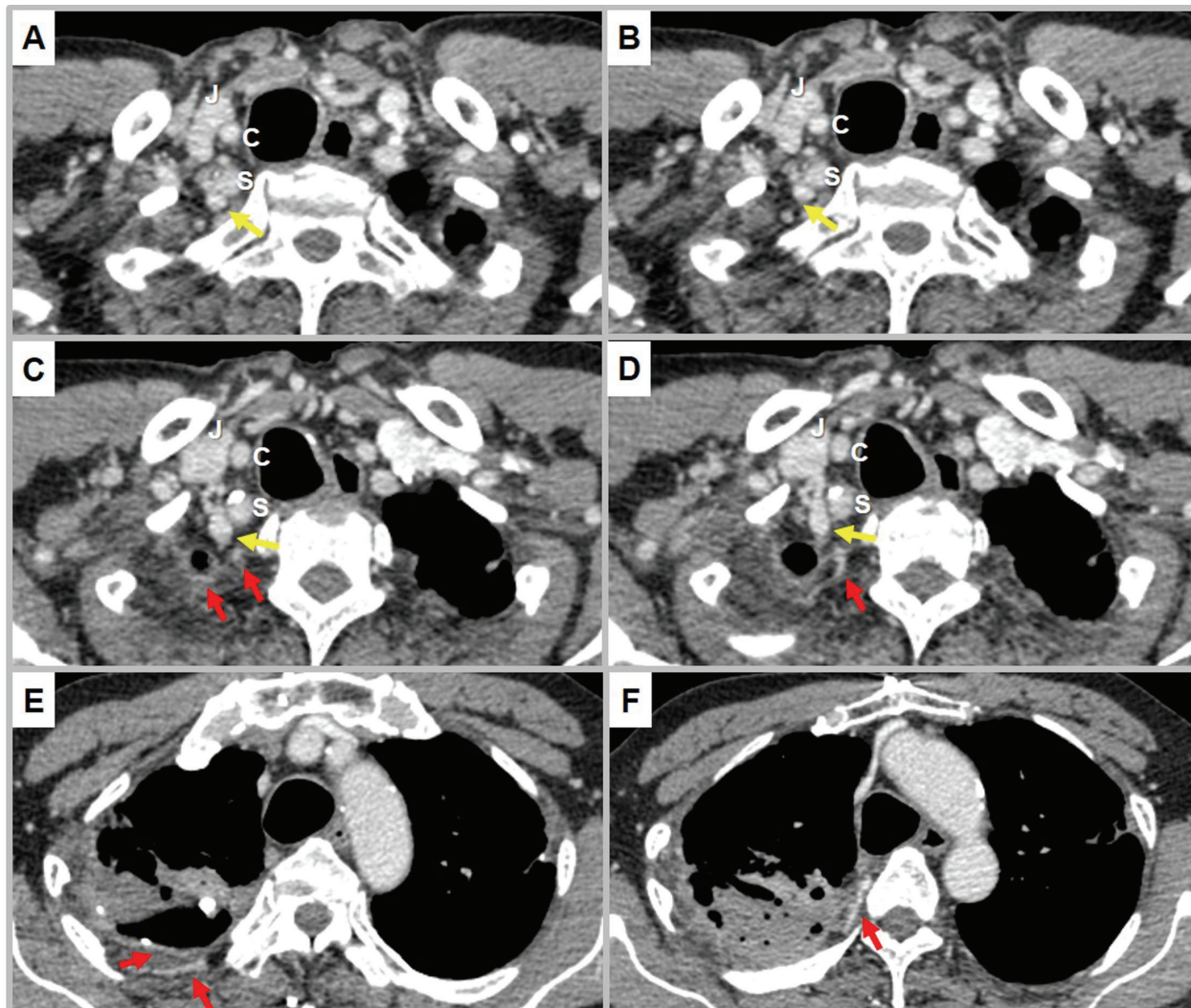


**Fig. 3.** Embolization of the bronchial arteries. After super-selection of the bronchial arteries, angiography revealed enlarged and tortuous small arterial branches supplying the right upper lobe (A) and superior segment of the right lower lobe (B), with marked hypervascularity of the affected lung (C). The hypervascular area became invisible after successful embolization with gelfoam cubes (D).

CT (Figure 4) and in the thoracic angiography (Figure 2B and 2C) until rebleeding occurred. To date, there is no consensus regarding a thorough evaluation for potential NBSAs during a BAE, as the prevalence of bleeding from the NBSAs is low, and these procedures inevitably require a higher dose of contrast agent and radiation exposure, as well as prolonged procedure time. However, chronic lung disease with vascular enhancement within the pleural thickening should be considered as a red flag, and the primary treatment team should be aware of

the possibility of bleeding from an NBSA when planning the treatment approach.

In the present case, bronchoscopy was not used as a diagnostic tool because of the compromised cardiopulmonary reserve and the tentative bleeding site in the RUL. The use of bronchoscopy as an initial diagnostic tool has been challenged recently. For identification of bleeding sites, the effectiveness of bronchoscopy was higher in patients with moderate (64%) to severe (67%) hemoptysis than in those with mild hemoptysis (49%) [14]. However, the



**Fig. 4.** Review of contrast multidetector computed tomography images. A non-bronchial systemic artery (yellow arrow) originated from the right subclavian artery (S) at the lower neck level, extending downward to the thickening pleura of the right apex (A to D; J, right internal jugular vein; C, right common carotid artery). In addition, many enlarged arterial branches were found in the pleural thickening of the affected lung (red arrows, C to F).

more severe the hemoptysis, the more difficult it is to visualize the airway, and the higher the risk of the patient being unable to tolerate the procedure. Moreover, the procedure itself might induce mucosal irritation and recurrent bleeding [15]. The diagnostic yield of bleeding sites with bronchoscopy (73-93%) was reportedly lower than that with MDCT (63-100%) [5,8,15-16]. Bronchoscopy can be considered for use in

distinguishing tumors from blood clots in the airways, for simultaneous sampling for tissue specimens or microbial cultures, or for therapeutic interventions, such as single lung intubation, cold saline irrigation, and balloon inflation [4,6,8]. In addition to identifying bleeding sites, MDCT provides information regarding an underlying lung disease, such as bronchiectasis, lung cancer and pulmonary infection [6,8].

Combined assessment with MDCT and bronchoscopy was reported to further improve diagnostic accuracy [14,17]. With a specific contrast protocol, MDCTA might provide more detailed thoracic vascular mapping, including BAs and NBSAs, which would be superior to conventional angiography [18]. A pre-embolization MDCTA could reduce the rates of rebleeding and the need for urgent surgical resection [8,19]. MDCTA should be considered as the first-line diagnostic tool in the management of massive hemoptysis [7-8].

The treatment modality chosen may also have a great impact on the outcome. In general, the use of BAE as a first-line hemostatic intervention was associated with a high immediate control rate (>80%) [7-8]. However, rebleeding, which requires further definitive treatment such as surgical resection of the affected lungs to prevent recurrence, was noted in 10-33% of cases [4,7,9]. Urgent surgery to control massive hemoptysis was associated traditionally with high mortality (25-40%) [4-5,10-11]. However, recent studies have found that surgical outcomes could be improved to approximately the same level as those in elective surgeries by achieving temporary control of bleeding preoperatively, with either BAE [20-21] or rigid bronchoscopy [21-22]. Patients in a PTB endemic area with massive hemoptysis after an emergency BAE might benefit from a subsequent surgical intervention, especially in the presence of high risk factors such as the absence of active PTB, the presence of aspergilloma, a need for blood transfusion, and an incomplete cessation of hemoptysis [23]. Our case was comparable to this risk-stratification. In addition, an increasing amount of evidence has shown that ECMO can effectively support patients with hemoptysis-related airway obstruction until a definite hemo-

static intervention is carried out [24].

The long-term outcomes of BAE are not promising the recurrence rate after BAE is 10-60% [7,9], and recurrence was associated with high mortality [25]. Late recurrence usually results from recanalization of the embolized vessels, angiogenesis and vascular recruitment, particularly in patients with advanced PTB, bronchiectasis, aspergilloma, and lung cancers [7,9]. Massive hemoptysis in patients with chronic parenchymal lung disease warrants a comprehensive image review and an aggressive treatment plan.

In conclusion, management of massive hemoptysis requires a multidisciplinary approach. Initial treatment includes stabilization, securing the airway, workups for identification of bleeding sites, and nonsurgical temporary hemostasis. An MDCTA is considered the first choice for use in diagnosis, followed by systematic examination of all potential bleeding sources. Nevertheless, BAE serves as the first-line intervention for hemostasis. Early surgical resection after temporary control of bleeding leads to beneficial outcomes in selected cases with high rebleeding rates.

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## 大量咳血：不要忽視非支氣管動脈之體循環動脈 —病例報告及文獻回顧

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大量咳血是一種致命的急症。造成出血的來源主要是支氣管動脈（90%），少數會來自肺動脈（5%）或非支氣管動脈之體循環動脈。第一線止血措施通常是支氣管動脈栓塞，但再出血率仍然偏高，而手術切除病灶才是根除性的治療。源自非支氣管動脈之體循環動脈的出血經常被忽略，因而導致治療失敗。我們報導一位因陳舊性肺結核毀損的右上肺葉出血所導致大量咳血的病例，該出血之肺葉具有少見之雙重出血來源 - 包括支氣管動脈及非支氣管動脈之體循環動脈。雖然該案例之急性出血在接受支氣管動脈栓塞術後暫時止血，兩天後又再次大咳血導致心跳一度停止。在葉克膜的支持之下，病患先接受了右上肺葉切除術，但隨即又因來自右鎖骨下動脈的分支動脈出血再次接受緊急手術。病患雖然撐過急性出血期，最後仍死於反覆肺炎。詳盡的影像學評估、辨識容易再出血的影像學徵象與危險因子，以及妥善計畫各種治療方式為改善預後的關鍵。( *胸腔醫學* 2018; 33: 130-138)

關鍵詞：支氣管動脈栓塞，大量咳血，多偵測器電腦斷層，非支氣管動脈之體循環動脈

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