

Case Report

# Effective Strategies for Managing Sudden Hemoptysis Caused by Aorto-Bronchial Fistula during Cardiopulmonary Bypass: A Case Report

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## Abstract

Aorto-bronchial fistula (ABF) is a rare but life-threatening complication that can occur after thoracic endovascular aortic repair (TEVAR). The ABF clinical diagnosis can be challenging due to its insidious symptoms and potential for misdiagnosis. Managing endobronchial hemoptysis caused by ABF during cardiopulmonary bypass (CPB) is challenging due to limited clinical experience. We present a case of a patient who was previously treated with TEVAR for a thoracic aortic dissection and endovascular abdominal aortic aneurysm repair for an abdominal aortic aneurysm. The patient was admitted with intermittent hemoptysis over 1 year and chest pain for 3 days. Aortic computed tomography angiography (CTA) showed a recurrent dissection of the aortic arch. We encountered endotracheal hemoptysis during total arch replacement combined with a stented frozen elephant trunk under CPB. Due to the patient's prior history of TEVAR, the ABF was eventually diagnosed during the procedure; however, with the implementation of a series of measures, we were able to successfully resuscitate the patient. The literature suggests that this may be an exceedingly rare case of ABF successfully treated during CPB. Currently, there are no established clinical guidelines or consensus for the diagnosis and treatment of ABF after TEVAR due to the lack of case reports with extensive data. Timely identification of the bleeding bronchus, early activation of blood cell salvage, early neutralization of heparin activity, and timely resection of the diseased lobe are key to treating patients with ABF during CPB.

## Keywords

aorto-bronchial fistula; TEVAR; cardiopulmonary bypass; hemoptysis; case report

## Introduction

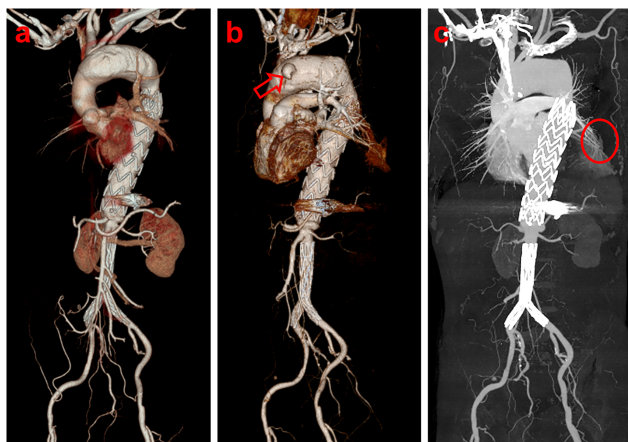
Aorto-bronchial fistula (ABF) is a rare and life-threatening complication that occurs after thoracic endovas-

cular aortic repair (TEVAR) and leads to hemoptysis [1]. If not promptly addressed, hemoptysis can progress to hemorrhagic shock and potentially result in death. ABF is a complex condition that can be misdiagnosed. Prompt repair of postoperative TEVAR endoleaks is crucial. Previous systematic reviews have indicated that endovascular repair is a viable option with favorable short-term outcomes [2,3]. However, there is limited evidence regarding the long-term outcomes of endovascular repair. Previous case reports have described successful treatment of this condition using open surgical techniques such as ascending aorta-descending aorta bypass, aorta-subclavian artery bypass, and subsequent resection of the descending aorta [4]. A literature review suggests considering a composite procedure, which involves interventional repair of the aorta accompanied by a pneumonectomy or a direct repair of the involved bronchus followed by coverage with a muscular or pleural flap, along with broad-spectrum intravenous antibiotic therapy [5]. Due to the rarity of ABF, most reports are based on small case studies or individual cases, which makes it challenging to establish treatment guidelines or a consensus for its management. In this report, we present a case of a patient with an aortic arch dissection who underwent total arch replacement combined with a stented frozen elephant trunk with the assistance of cardiopulmonary bypass (CPB). During the surgery, the patient developed massive hemoptysis. Given the patient's history of a previous TEVAR, we suspected the presence of a postoperative ABF. Fortunately, we were able to effectively manage this complication and review its management in this case report.

## Case Presentation

A 56-year-old male was admitted to the hospital with intermittent hemoptysis, chest tightness that started a year ago, and chest pain for 3 days. He had a long history of hypertension and experienced a descending aortic dissection five years ago. He underwent A TEVAR covered the descending aorta and made an uneventful recovery. Three years later, the patient experienced an abdominal aortic pseudoaneurysm and underwent successful endovascular abdominal aortic aneurysm repair with branched graft.

However, his blood pressure remained uncontrolled. He had no family history of genetic disorders or aneurysmal disease. The patient's condition continued to deteriorate, and due to uncontrolled hypertension, he developed an aortic arch aneurysm with a dissection. Aortic computed tomography angiography (CTA) scan showed a hematoma in the repaired descending aorta with an aortic arch dissection (Fig. 1); segmental atelectasis with infection in the left lower lobe and a small pleural effusion (Fig. 2b,c). The abdominal aorta showed only post-interventional changes (Fig. 1, Fig. 2a). He was admitted to the hospital with a plan to perform a total arch replacement combined with a stented frozen elephant trunk. We suspected that the hemoptysis might be related to bronchial artery involvement. However, no significant bronchial artery or ABF was observed on the aortic CTA (Fig. 1c), so we hypothesized that it might be related to the left lower lung atelectasis and concomitant infection. After admission, all cultures were negative and the blood pressure was maintained below 120/60 mmHg.



**Fig. 1. Aortic CTA in 3D.** (a) The CTA schematic image of the patient taken two years ago shows dilatation of the aortic arch with post-stent implantation status. (b) The current CTA after hospitalization reveals the formation of an aortic arch dissection (red arrow). (c) No obvious bronchial arteries and ABF were observed on CTA (red circle). CTA, computed tomography angiography; ABF, aorto-bronchial fistula; 3D, Three-dimensional.

Preoperative transesophageal echocardiography showed septal hypertrophy. We established CPB using the right atrium and femoral artery and brachiocephalic trunk. Subsequently, the ascending aorta and aortic arch were opened on deep hypothermic low-flow CPB, revealing the dilatation of the ascending aorta and aortic arch and the formation of a dissection flap in the aortic arch. After resection of the thrombus within the dissection, a 28-mm descending aortic stent graft with a left subclavian artery branch was implanted (Fontus Branching Stent Graft System, MicroPort Endovascular MedTech, Shanghai, China). The proximal aortic arch was anastomosed with a

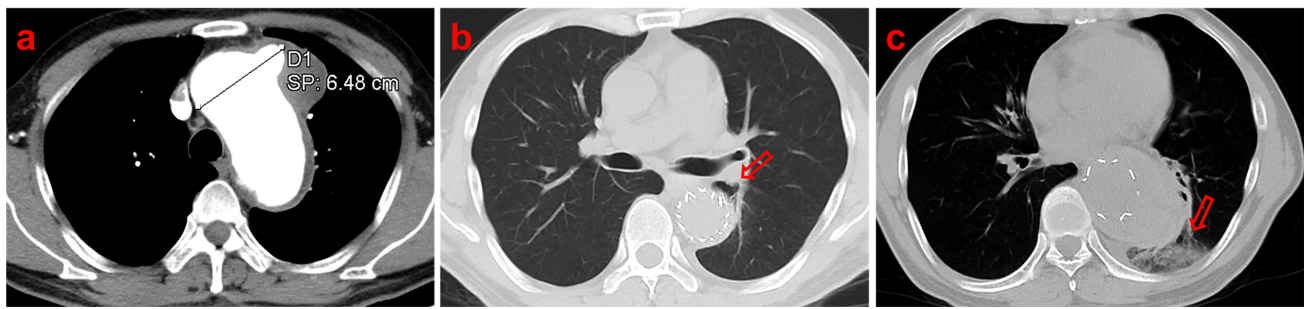
28-mm four-branch stent (Maquet, Hechingen, Germany). After systemic circulation was restored, the left carotid and brachiocephalic trunk artery were anastomosed to the graft and reinforced with intermittent mattress sutures (Fig. 3a). Massive intraoperative airway bleeding was then observed. Suction and exploration using a fiberoptic bronchoscope failed to identify the bleeding tracheal site. The original tracheal tube was immediately replaced with a double-lumen tube, and it was noted that the hemoptysis originated from the left trachea. We reversed the heparin with protamine sulfate and gradually weaned from CPB. The airway bleeding decreased slightly but still persisted. Upon opening the left pleura, pulmonary atelectasis were noted in the left lower lung. An additional left anterolateral incision in the fifth intercostal space was made and further dissection revealed that the left lower lung was tightly adherent to the descending aorta, forming an ABF. A left lower lobectomy was performed and the hemoptysis was significantly reduced (Fig. 3b,c). The fistula from the aorta was repaired and covered with a dacron patch and the bleeding site was compressed with hemostatic gauze using a porcine fibrin sealant kit (Fig. 3c). The intraoperative blood loss was approximately 20,000 mL.

Pathologic findings in the aortic arch vessels revealed mucous degeneration of the arterial wall tissue, partial detachment of the arterial intima, and the presence of a mixed thrombus, all consistent with a diagnosis of aortic dissection (Fig. 4a). Examination of the lung tissue showed extensive congestion and edema in the alveolar cavities, deposition of hemosiderin and histiocytic reaction, destruction of some alveolar structures, and proliferation of fibrous tissue (Fig. 4b). Following the surgery, the patient's vital signs remained stable in the intensive care unit (ICU). However, there was a slight impairment in oxygenation, with an oxygenation index ranging between 100–150. A postoperative chest computed tomography (CT) scan revealed atelectasis in the left lung, bilateral lung infiltrates, and a pleural effusion. With aggressive treatment, both lungs showed significant improvement, and the oxygenation index returned to normal (Fig. 5). Ten days after the surgery, the patient was successfully weaned off ventilator support and transferred out of the ICU. He now requires regular follow-up appointments and his long-term prognosis is being closely monitored.

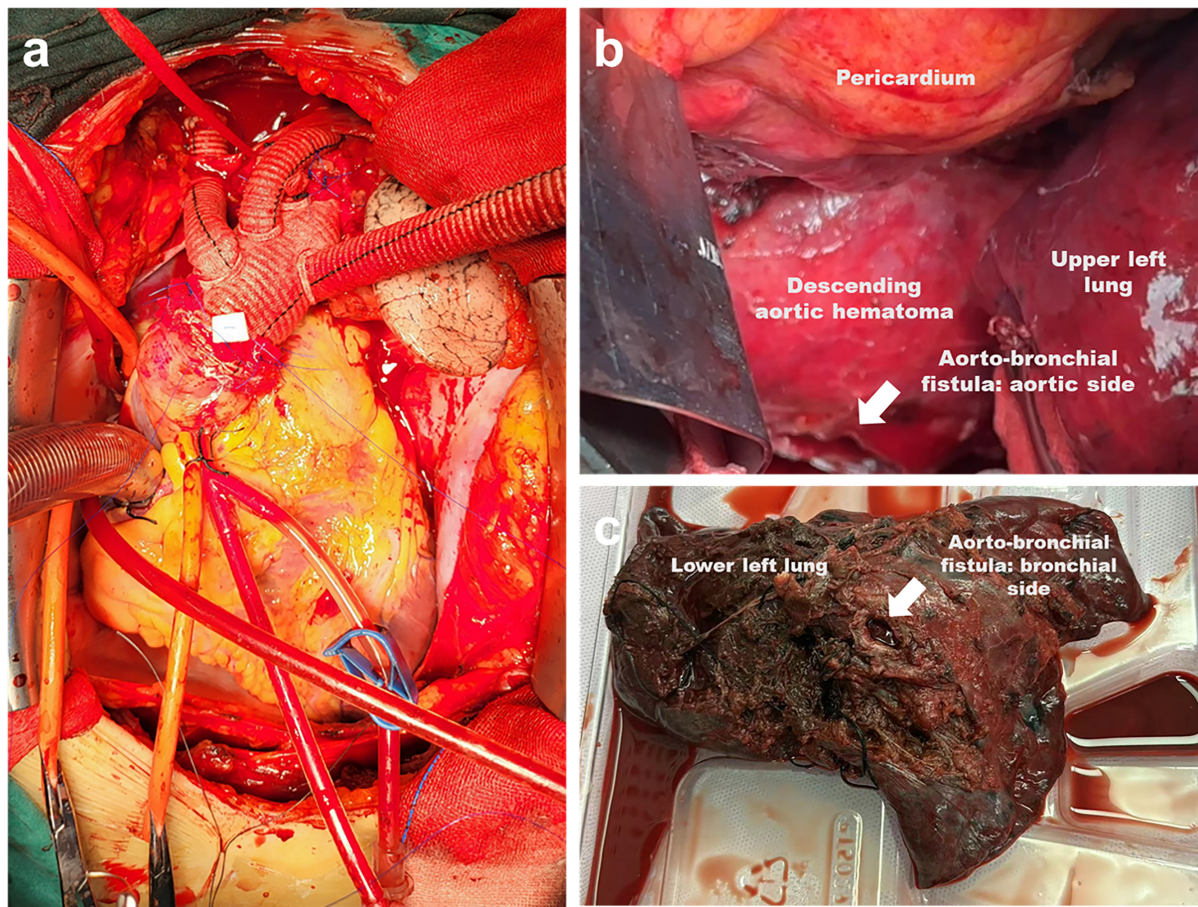
## Discussion

This patient initially presented with an aortic dissection involving the aortic arch. The patient had a history of recurrent aortic dilation, ruling out diagnoses such as immune system disorders like Takayasu arteritis or giant cell arteritis. Although the patient had a history of hemoptysis, there was no recurrence of hemoptysis during this hospitalization after the blood pressure was well controlled. There-





**Fig. 2. 2D slices of aortic CTA and chest CT.** (a) CTA schematic showing aortic arch dilatation and dissection (6.48 cm at widest point). (b) Chest CT showing suspected ABF in the descending aorta (lung windows, red arrow). (c) CT shows signs of atelectasis and infection in the lower left lung (mediastinal windows, red arrow). 2D, Two-dimensional; CT, Computed Tomography.

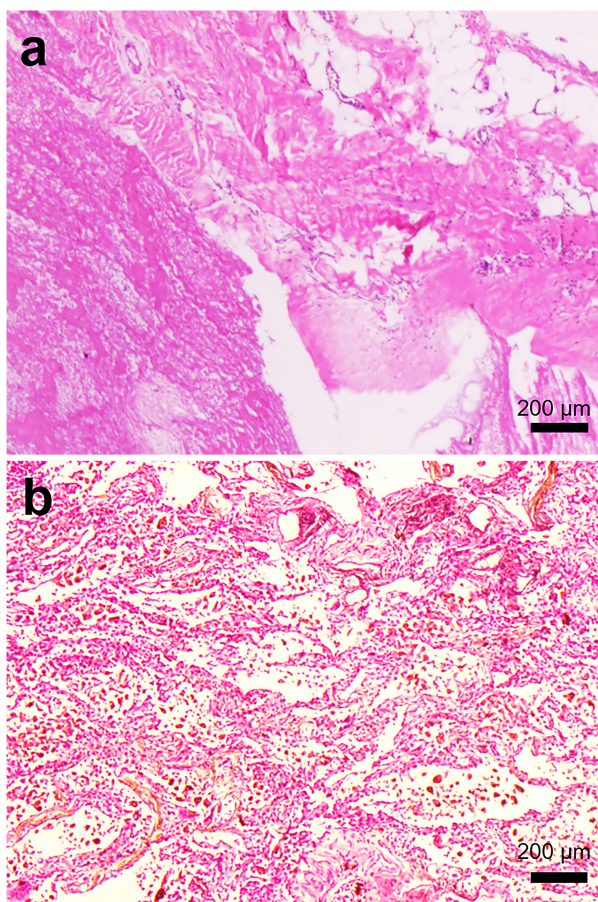


**Fig. 3. Intraoperative diagram.** (a) Status after total arch replacement combined with stented elephant trunk implantation (Pre-pulmonary hemorrhage). (b) Descending aorta after lobectomy, with the stump on the descending aortic side of the ABF visible (white arrow). (c) Resection of the left lower lung, bronchus side of the ABF is visible (white arrow).

fore, we overlooked the possibility of an existing ABF. Despite the use of different types of imaging such as bronchoscopy, chest X-ray, CT scan, and echocardiography, the diagnosis can still be challenging [6]. Bronchoscopy is the most sensitive and specific method for diagnosing ABF, but it usually requires sedation and carries a risk of thrombus dislodgement, which can lead to fatal bleeding [7]. We did not use bronchoscopy preoperatively because he suffered

from an aortic dissection. During the descending aortic arch replacement and frozen elephant trunk stent implantation under CPB, the patient experienced hemoptysis approximately 4 hours after heparinization towards the end of the surgery. We considered several potential factors contributing to this complication: (i) The implanted descending aortic stent may have caused the displacement or mechanical changes in the original stent. (ii) Heparinization may have





**Fig. 4. Histological sections.** (a) Haematoxylin-eosin-stained tissue sections of aortic arch vessels. (b) Haematoxylin-eosin-stained tissue sections of congested lung tissue. Scale bar = 200  $\mu$ m.

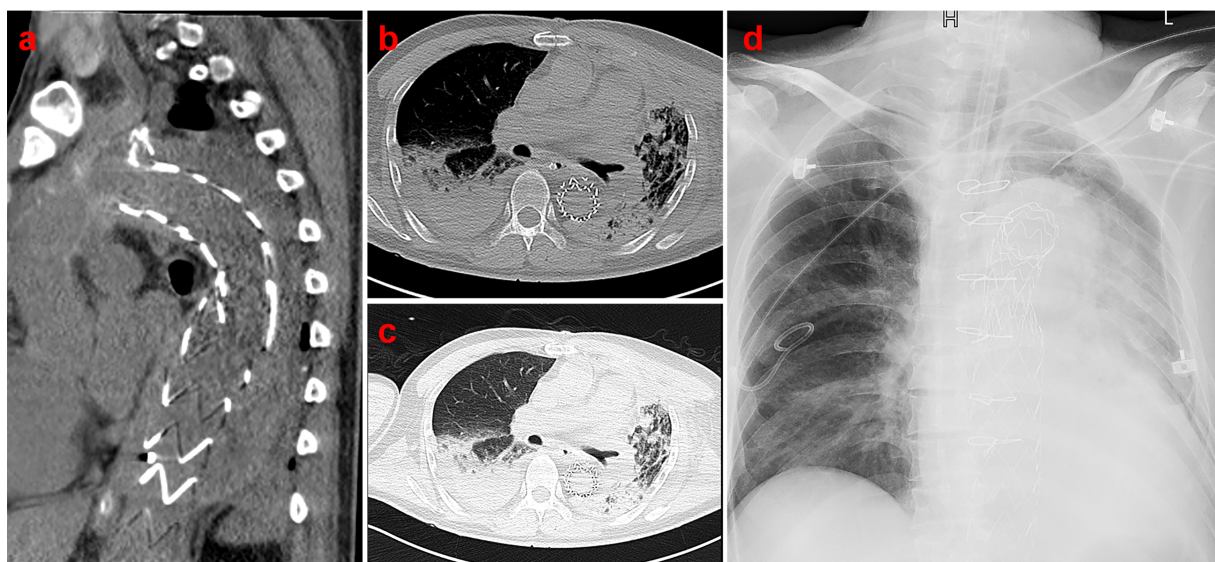
facilitated the dislodgement of pre-existing thrombi. (iii) Hemoptysis occurred shortly after the restoration of circulation through the femoral artery, which may be related to the changes in blood pressure following the resumption of circulation.

ABF is a rare clinical case that refers to the presence of anomalous connection between the aorta and the bronchus. According to the European Register of Complications of TEVAR, the incidence of ABF after TEVAR is approximately 0.56% [8]. Before 1960, infectious diseases (such as tuberculosis, syphilis) and fungal aneurysms were the primary causes of aneurysms leading to ABF. However, in the subsequent two decades, atherosclerosis and medical diseases became the predominant etiologies. As TEVAR continues to advance in the management of descending aortic disease, there has been a growing number of reported cases of ABF associated with endovascular repair. Intermittent hemoptysis is a frequently observed symptom that prompts patients to seek medical help. Hypovolemic shock was found to be present in of 6.46% of patients. Additional clinical signs and symptoms include chest or back pain, cough,

and difficulty breathing [9]. At present, treatment for ABF includes conservative management, open surgical intervention, and endovascular repair. Even though ABF is relatively uncommon, its treatment presents significant challenges and the prognosis is typically unfavorable, particularly when managed conservatively with antibiotics alone [10]. Endovascular repair of the aortic aneurysm has shown good short-term results and is a feasible alternative approach [3]. Nevertheless, the long-term efficacy and safety of these approaches remain unknown. Although TEVAR can rapidly control bleeding with minimal invasiveness, it fails to address the root cause and does not resolve any communication with the respiratory tract, which increases the risk of recurrent ABF. Currently, De Rango *et al.* [11] and Hu *et al.* [12] lean towards open surgery rather than endovascular repair as the preferred treatment for ABF. Surgical procedures using in situ prosthetic graft replacement and simultaneously through extra-anatomic bypass have been employed [4]. In situ graft replacement surgery is an effective option for small fistulas and patients with controlled systemic infection. The advantage of in situ surgery is that both the bronchus and the aorta can be repaired in a single procedure. On the other hand, in situations where a complex fistula with severe infection and adhesions is present, extra-anatomic bypass surgery is favored to reduce the likelihood of recurring infections. In this context, De Rango *et al.* [11] propose that open surgery might be a safer choice, particularly due to the frequent requirement for repeat TEVAR. Open surgery offers advantages over endovascular repair, as it enables thorough debridement, aortic graft replacement, and coverage of the stump, thereby reducing the risk of recurrent infection. However, there is no consensus in the literature regarding the optimal approach for treating ABF, and larger studies are required to clarify this issue. An effective strategy for severely unstable patients may be to use a stent to control bleeding in an emergency situation, followed by a delayed, more thorough open repair once the patient is stabilized [13].

There have been reported cases of patient deaths resulting from the inability to discontinue CPB due to ABF [9]. Our case illustrates the important management strategies in dealing with hemoptysis from ABF during CPB. (i) Promptly locate the source of bleeding in the trachea. We first used a bronchoscope to examine the trachea, but due to the presence of blood throughout the trachea, we could not identify the exact bleeding site. Therefore, we immediately placed a double-lumen endotracheal tube and observed a large amount of blood arising from only the left bronchus. (ii) Immediately initiate blood cell salvage for rapid transfusion to maintain blood pressure, although potential contamination is a concern. (iii) Neutralize heparin as soon as all graft anastomoses have been completed. (iv) Early resection of the left lower lung and identification of the bleeding site in the aorta. After administering hemostatic agents such as clotting factors, aminocaproic acid, snake venom





**Fig. 5. Postoperative diagram.** (a) Postoperative follow-up chest CT, showing the connection of our implanted descending aortic stent with branches (Fontus) shadow to the original stent. Chest CT image at 3 days postoperatively, (b) with bilateral stenting visible in the mediastinal windows and (c) pulmonary exudates and foci of infection visible in the lung windows. (d) Chest X-ray on the eighth postoperative day showed that the right lung was back to normal, the ventilation of the upper left lung had improved compared to the previous day.

thrombin-like enzymes, and fibrinogen, the bleeding significantly improved. Subsequently, mechanical compression was applied, leading to the cessation of bleeding. We then used a dacron patch to suture the aorta and applied biological glue for compression to achieve temporary hemostasis. These interventions have some limitations, as their still may be risk of rebleeding from the diseased aorta. Possible complications include postoperative hypoxia and the potential for mediastinal infection. Our primary focus is on temporary treatment, and we plan to perform a second-stage surgery after the patient recovers from this operation. We are still discussing the subsequent treatment plan.

Based on this case, we have summarized the following lessons learned: (i) For patients with aortic dissection, there is a high risk associated with bronchoscopy examination. In this case, the CTA examination did not detect the contrast agent entering the bronchus, leading to the failure to diagnose the ABF before surgery. The clinical presentation of ABF can be subtle and difficult to diagnose. (ii) In this case, the initial symptoms of hemoptysis, lung collapse, and infection were initially thought to be related to bronchial artery rupture, possibly caused by hypertension or aortic dissection. (iii) It may have been more reasonable to include an aortic arch replacement, combined with descending aortic prosthetic vascularization and left lower lobectomy, if the ABF could have been diagnosed before the operation. A previously reported case of aorto-bronchial fistula after TEVAR resulted in hemoptysis requiring urgent repair, and CTA showed an aorto-bronchial fistula and type Ia endoleak [14]. Providing proximal closure and left common carotid

artery and subclavian artery revascularization through the use of a fenestrated endograft also provides a therapeutic option of total endoluminal repair in the treatment of this patient, but the patient still faces the therapeutic risks of ABF recurrence and infection in the long term if this option is used [15].

## Conclusions

In conclusion, ABF is a rare complication of TEVAR that results in a high mortality rate and poor prognosis during cardiopulmonary bypass. ABF needs to be considered when a large amount of bronchial hemoptysis combined with a previous history of TEVAR is encountered during surgery. Intraoperatively, airway management, blood replacement, and cardiopulmonary bypass management, are crucial for a favorable outcome. Double lumen endotracheal intubation not only has the advantage of preserving single lung ventilation oxygenation and facilitating CPB but also helps to define the bleeding bronchial branches. Prompt resection of the affected lung parenchyma and rapid neutralization of heparin help to reduce bleeding. The CARE checklist was used when writing this case report (Supplementary Table 1).

## Availability of Data and Materials

All data generated or analyzed during this study are included in this manuscript.

## Author Contributions

XL, XH and SY designed the research study. KW performed the research and drafted the initial manuscript. CY analyzed the data. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript. All authors have participated sufficiently in the work to take public responsibility for appropriate portions of the content and agreed to be accountable for all aspects of the work in ensuring that questions related to its accuracy or integrity.

## Ethics Approval and Consent to Participate

No IRB approval was required for this study. The Affiliated Hospital of Guizhou Medical University IRB deems case reports of less than three patients not to constitute human subject research and therefore not to require IRB review and approval. Written informed consent was obtained from the families of the patient for scientific activity including publication of this case report.

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## Conflict of Interest

The authors declare no conflict of interest.

## Supplementary Material

Supplementary material associated with this article can be found, in the online version, at <https://doi.org/10.59958/hsf.6889>.

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