Bronchial Artery Embolization for Hemoptysis: Two Cases of Primary Bronchial Artery Racemose Hemangioma

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Each author contributed to the idea of the paper, the research of the case, and the editing of the case report.

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ABSTRACT

We report two cases of primary bronchial artery racemose hemangioma (RHBA) in female patients presenting with hemoptysis as the dominant manifestation complicated by hemodynamic instability. Following a multidisciplinary team (MDT) consensus, bronchial artery embolization (BAE) was implemented as the primary intervention. Procedural success was achieved in both cases, resulting in the complete resolution of hemoptysis and hemodynamic stabilization. Postprocedural surveillance demonstrated sustained efficacy without recurrence during the follow-up period. This study validated BAE as an effective and safe first-line therapeutic approach for RHBA while further investigating the correlation between the embolization strategy and long-term clinical outcomes.

CASE REPORT

BACKGROUND

Primary bronchial artery racemose hemangioma (RHBA) is a rare vascular malformation characterized by segmental ectasia, tortuosity, and plexiform proliferation of the bronchial arteries. Affected individuals may manifest respiratory symptoms and rupture may trigger massive hemoptysis, inducing chest tightness and dyspnea. Consequently, early detection and intervention are imperative to avoid life-threatening hemorrhage and, with refinements in endovascular techniques, BAE should be considered as the first-line treatment for RHBA [1]. However, standardized embolization protocols have not been established yet. This case series delineates two primary RHBA presentations, aiming to elucidate the critical diagnostic and therapeutic paradigms, heighten clinical vigilance among practitioners, and inform evidence-based management decisions.

CASE PRESENTAION

Case 1

A 47-year-old woman presented with hemoptysis, chest tightness, and pleurodynia in May 2025. Upon admission, a clinical evaluation revealed hypotension (blood pressure, 92/58 mmHg), tachycardia (heart rate, 116 beats/min), hypoxemia (oxygen saturation, 92%), and anemia (hemoglobin concentration, 103g/L). Contrast-enhanced thoracic computed tomography (CT) revealed the presence of a mediastinal hematoma alongside a long-segment, tortuous, dilated, and plexiform right bronchial artery located at the level of the T6 vertebra (Figure 1A). The patient subsequently developed hemodynamic instability, manifesting as a precipitous decline in blood pressure (84/50mmHg) and hemoglobin (80g/L), consistent with hemorrhagic shock. Following MDT consultation, digital subtraction angiography (DSA) was performed for definitive identification of the bleeding source. Under local anesthesia, percutaneous femoral arterial access was obtained. Thoracic aortography utilizing a Cobra catheter delineated the characteristic angiographic findings of a tortuous, dilated, plexiform bronchial artery (Figure 1B), confirming the RHBA diagnosis. Subsequent BAE was performed under DSA guidance. Initial embolization was achieved using two detachable coils (Cook Medical, Inc., Bloomington, USA; IMWCE-35-5-5MREYE) with documented cessation of antegrade flow. Subsequent thoracic aortography using a pigtail catheter identified two additional bronchial arteries supplying the hemorrhagic region of the lungs (Figure 1C). These vessels were selectively cannulated using a Simmons catheter with superselective microcatheter confirmation, followed by embolization of the target vessels. Six additional coils of varying diameters (Cook Medical, Inc., Bloomington, USA; MWCE-18S-3/2-TORNADO and MWCE-18S-4/2-TORNADO) were deployed to occlude the communicating branches and proximal segments of the culprit vessels (Figures 1D,1E). Post-embolization angiography confirmed successful "stump-like" occlusion of the target arteries (Figure 1F). The postoperative course was unremarkable, and no fever or pleurodynia was observed. Her hemoglobin levels progressively recovered to 106 g/dL. Follow-up non-contrast thoracic CT12 days post-procedure demonstrated significant resolution of the previously noted bilateral pulmonary patchy opacities, a marked reduction in right pleural effusion volume, and improvement in right lower lobe atelectasis (Figure 1G). The patient remained recurrence-free at the 3-month follow-up.

Case 2

A 28-year-old woman presented with massive hemoptysis (estimated volume: 400 mL) in September 2024. Physical examination revealed an acutely ill condition, characterized by coarse bilateral breath sounds and moist crackles localized to the right lower lobe. Contrast-enhanced thoracic computed tomography revealed a tortuous, dilated, and plexiform vascular structure originating from the thoracic aorta, consistent with hypertrophy of the right bronchial artery (Figure 2A). These imaging features were highly suggestive of RHBA. Following MDT assessment, BAE was performed. Under local anesthesia, selective catheterization of the right bronchial artery ostium was performed using a 5F Cobra catheter under fluoroscopic guidance with contrast injection. Angiography revealed significant arterial hypertrophy, tortuosity, and disorganization with a distal plexiform vascular network and active contrast extravasation, indicative of vessel rupture (Figure 2B). This angiographic confirmation, in conjunction with the CT findings, established a definitive diagnosis of RHBA; superselective catheterization of the culprit vessel was performed. Using a flow-directed technique, four detachable microcoils (Cook Medical, Inc., Bloomington, IN, USA; MWCE-18S-4/2-TORNADO) were deployed within the distal segment of the pathological vessel. Postembolization angiography confirmed successful occlusion with complete cessation of contrast extravasation (Figure 2C). The postoperative course was uneventful, with immediate resolution of the hemoptysis and no complications. The patient was discharged on day 10 of hospitalization. At the 5-month follow-up contrast-enhanced thoracic CT demonstrated complete resolution of the previously noted bilateral pulmonary parenchymal opacities and punctate hyperdensity in the right hilum. Re-expansion of the right middle and lower lobes was satisfactory (Figure 2D). The patient remained asymptomatic with no evidence of recurrence during the 11-month follow-up period.

DISCUSSION

The RHBA was first reported by Babo et al.[2] in 1976. The infrequency of this condition prevents the establishment of formal diagnostic criteria based on the limited number of documented cases, although no universally standardized diagnostic algorithm exists, characteristic features proposed by Japanese investigators and supported by literature consensus include recurrent hemoptysis, long-segment arterial tortuosity, dilatation, and hyperplasia demonstrated by contrast-enhanced CT or DSA, and potential association with bronchial artery aneurysms or plexiform vascular complexes [3]. The predominant clinical manifestations of RHBA include hemoptysis, cough, chest pain, chest tightness, and dyspnea. Notably, some patients with RHBA may remain asymptomatic, and no correlation has been found between size and risk of rupture [4]. Therefore, even in asymptomatic patients, immediate intervention is warranted upon diagnosis, which leads to high clinical suspicion in patients presenting with unexplained hemoptysis or hemorrhagic pleural effusion concomitant with bronchial abnormalities. Angiography of the bronchial arteries is currently essential for the diagnosis [5]. By injecting a contrast agent, direct signs of the responsible vessel include contrast agent overflow, and indirect signs include irregular expansion and tortuosity of the main supply artery and an increase and disorder of vascular branches in the lesion area. DSA can also provide guidance for BAE. Therefore, once RHBA is suspected, enhanced chest CT and DSA should be prioritized for auxiliary diagnosis [6].

BAE was initially implemented by Rémy et al. [7] in 1974 to manage hemoptysis. A systematic review conducted by Karlafti et al. [8]. demonstrated that BAE achieved a clinical success rate of 92.46% for the management of massive hemoptysis. Therefore, BAE is an effective treatment for RHBA [9]. This endovascular intervention offers significant clinical advantages, including minimal procedural trauma, accelerated postoperative recovery, shortened hospitalization duration, preservation of pulmonary functional capacity, and profound short-term therapeutic efficacy. Consequently, BAE should be considered a first-line treatment for RHBA [1,10], BAE uses a range of embolic materials, including N-butyl cyanoacrylate (NBCA), gelatin sponges (GS), and metallic coils. Coils are permanent embolic modalities used to treat cerebral aneurysms. Based on deployment mechanism, coils are categorized as either pushable or detachable. Data presented at the 2025 European Interventional Embolization Therapy Conference (ET2025) indicated no statistically significant difference in technical success rates between pushable (99.5%) and detachable (99.7%) coils. Due to their durability and capacity for rapid flow reduction, coils are widely utilized in clinical embolization procedures. Compared to other embolic materials, coil embolization for BAE is associated with a lower incidence of spinal cord infarction [11].

Currently, there is no standardized embolization strategy for treating RHBA. Proximal embolization targets the vasculature with a high- flow volume, enabling rapid hemostasis, which is particularly advantageous in hemodynamically unstable patients with massive hemoptysis. The typically large luminal diameter (>2 mm) of the proximal vessels accommodates permanent embolic materials, facilitating long-term occlusion. However, proximal embolization provides only transient flow interruption and predisposes patients to collateral vessel formation and eventual recanalization [12]. Nevertheless, this approach often fails to occlude distal aneurysm-feeding branches originating beyond the embolization site, resulting in incomplete devascularization. Furthermore, embolic material reflux may cause non-target embolization, potentially inducing catastrophic complications, such as esophageal necrosis, myocardial ischemia, and spinal cord injury [13]. Despite the dual blood supply to the bronchial wall, collateral circulation may be insufficient during pathological states, such as infection or malignancy. Under these circumstances, complete proximal flow occlusion predisposes patients to ischemic necrosis, mural perforation, and subsequent bronchopleural or bronchoesophageal fistula formation. Proximal embolization is contraindicated when the spinal artery originates in the bronchial trunk [14]. In Case 1, severe vascular tortuosity, dilation, and acute angulation precluded distal microcatheter advancement to the terminal vasculature. Published literature indicates that flow-directed coil deployment is a viable embolization strategy under such anatomical constraints [15]. Consequently, pushable coils were sequentially deployed to achieve proximal embolization of each culprit vessel, resulting in the complete cessation of hemoptysis. Although BAE is an effective palliative intervention, it has a reported recurrence rate of 21.2% [16]. Given the complex multi-source, multi-branch vascular architecture observed in Case 1, a meticulous technique was employed. Serial angiography ensured comprehensive identification of all feeding vessels, followed by definitive embolization to optimize therapeutic efficacy and mitigate recurrence risk. The BAE has significant clinical utility because of its high repeatability. It remains the preferred intervention for recurrent hemoptysis secondary to nonneoplastic vascular anomalies. Contemporary evidence shows that repeat BAE for recurrent hemoptysis following successful initial embolization is safe and efficacious [17]. Moreover, complication rates do not significantly increase; however, after proximal embolization, blood vessels become occluded, and the original vascular structure is unclear. During the second embolization, the possibility of increased difficulty in embolization may increase because of interruption of the path [18]. Conservative management or surgical lobectomy is a viable alternative in these patients. From a theoretical perspective, embolization efficacy is optimized when performed as distally as feasible within a bleeding artery. Distal embolization is often prioritized as the primary strategy [13], particularly when microcatheter access to the aneurysmal sac is safely and stably achieved. Distal superselective embolization can effectively block the blood supply to the terminal part of the aneurysm, directly acting on the aneurysm nidus and reducing damage to the surrounding normal tissues. Simultaneously, effective suppression of side branch recruitment reduces risk, and deployment distal to the origin of the spinal artery effectively prevents reflux into the anterior spinal arterial system [14]. However, in an actual operation, distal Superselective Embolization is not necessarily ideal when closer to the vascular terminal. The embolization material is affected by the impact of blood flow within the target vessel and usually remains at the vascular level corresponding to its diameter. If the catheter is inserted too deeply, it may cause catheter entrapment, completely blocking the blood flow in the target vessel, thereby causing the embolic agent to accumulate at the catheter end, which in turn affects the embolization effect. In contrast, distal Superselective Embolization requires the selective insertion of the microcatheter into the fine distal branches of the lesion. This not only places high demands on the vascular anatomical structure but also poses a significant challenge to the operator's technical proficiency. This technical challenge was exemplified in case 2, in which a solitary RHBA lesion was successfully managed through superselective microcatheter positioning and flow-directed coil deployment within the distal pathological vasculature. Research indicates that distal embolization is significantly superior to proximal embolization in terms of efficacy, safety, and long-term control rates.

This study documents successful and immediate clinical outcomes in two rare cases of RHBA managed with distinct BAE embolization strategies. While both interventions achieved immediate hemostatic control, longitudinal assessment of therapeutic durability requires extended follow-up surveillance.

TEACHING POINT

Bronchial Artery Embolization (BAE) has emerged as an effective and minimally invasive treatment for managing primary bronchial artery racemose hemangiomas (RHBA), a rare vascular anomaly often presenting with hemoptysis. In two cases discussed, BAE successfully halted bleeding and led to full hemodynamic stabilization, showcasing its role as a first-line therapy. The critical diagnostic features of RHBA include tortuous, dilated, and plexiform bronchial arteries, typically identified through contrast-enhanced thoracic CT and digital subtraction angiography (DSA). Given its high success rate and minimal invasiveness, BAE should be considered the primary treatment option for patients with RHBA, especially those with massive hemoptysis.

QUESTIONS

Question 1: What is the most effective first-line treatment for primary bronchial artery racemose hemangiomas (RHBA)?

A. Surgical resection
B. Bronchial artery embolization (BAE)(applies)
C. Chemotherapy
D. Antibiotics

Explanation: BAE is the preferred first-line treatment for RHBA due to its minimally invasive nature, high success rate in controlling hemoptysis, and ability to stabilize hemodynamics

Question 2: Which imaging modality is crucial for diagnosing RHBA and guiding embolization therapy?

A. X-ray

B. MRI

C. Contrast-enhanced thoracic CT and DSA(applies)

D. Ultrasound

Explanation: Both contrast-enhanced thoracic CT and digital subtraction angiography (DSA) are essential for identifying RHBA and for planning the embolization procedure

Question 3: What is a common clinical feature of patients with RHBA?

A. Asymptomatic presentation

B. Severe hemoptysis(applies)

C. Headache

D. Abdominal pain

Explanation: RHBA typically presents with massive hemoptysis, a condition that can cause severe respiratory distress and hemodynamic instability, requiring immediate intervention

Question 4: What complication can arise from the embolization of RHBA if not done carefully?

A. Pneumothorax

B. Spinal cord infarction(applies)

C. Bronchospasm

D. Liver failure

Explanation: While embolization is generally safe, improper techniques, particularly in proximal embolization, can risk non-target embolization leading to complications such as spinal cord infarction

Question 5: Which of the following is true regarding the recurrence of hemoptysis after BAE?

A. BAE guarantees no recurrence of hemoptysis.

B. The recurrence rate for hemoptysis after BAE is high, near 50%.

C. BAE is associated with a recurrence rate of approximately 21.2%.(apllies)

D. Recurrence of hemoptysis is only seen after surgical resection.

Explanation: Although BAE is highly effective, a recurrence rate of about 21.2% has been observed, highlighting the need for follow-up imaging and potential repeat interventions

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FIGURES

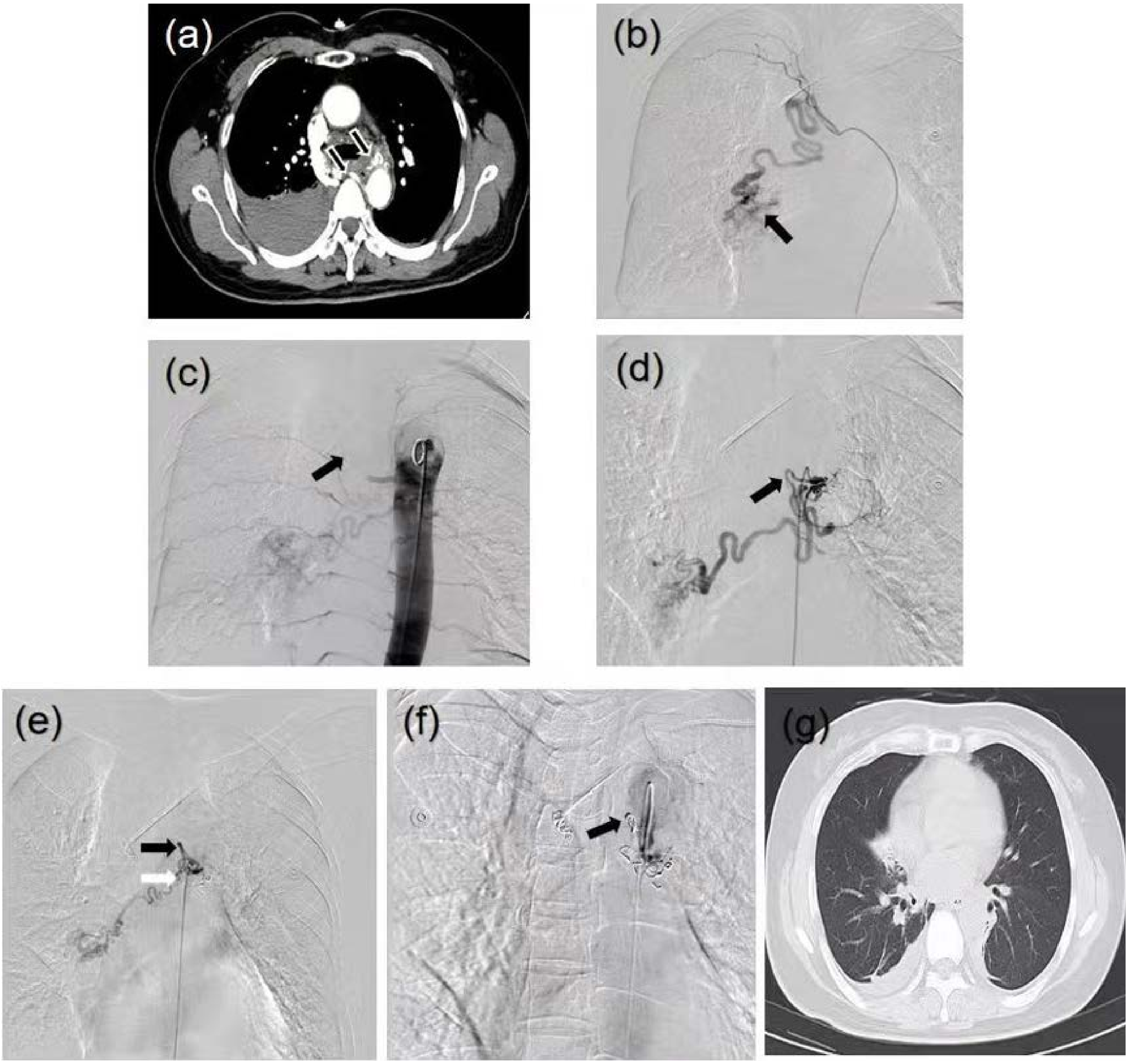

Figure 1: BAE treatment for RHBA. (A) Initial contrast-enhanced thoracic CT: Hyperdense right pleural effusion (41-58 HU) in posterior mediastinum and right hemithorax. Tortuous, dilated, and disorganized vascular structures in posterior mediastinum and right hilum. (B) DSA: Long-segment aneurysmal dilatation of right bronchial artery. Terminal contrast extravasation with disorganized vascular bed. (C) Post-embolization thoracic aortography (subtraction technique): Proximal coil embolization of superior RHBA. Two persistent hypertrophied bronchial arteries supplying hemorrhage territory. (D) Embolization of inferior RHBA: Proximal coil deployment with gelatin sponge augmentation. Visible communicating branch (arrowhead). (E) Post-communicating branch embolization: Coil occlusion of communicating branch (white arrow). Abnormal vascular blush in middle RHBA (black arrow). (F) Final embolization outcome: Proximal coil deployment in middle RHBA. "Stump-like" occlusion of all three RHBAs. Complete resolution of abnormal vascular blush.(G) 12-day follow-up non-contrast CT: Interval resolution of bilateral pulmonary opacities. Significant reduction in right pleural effusion. Improved re-expansion of right lower lobe.

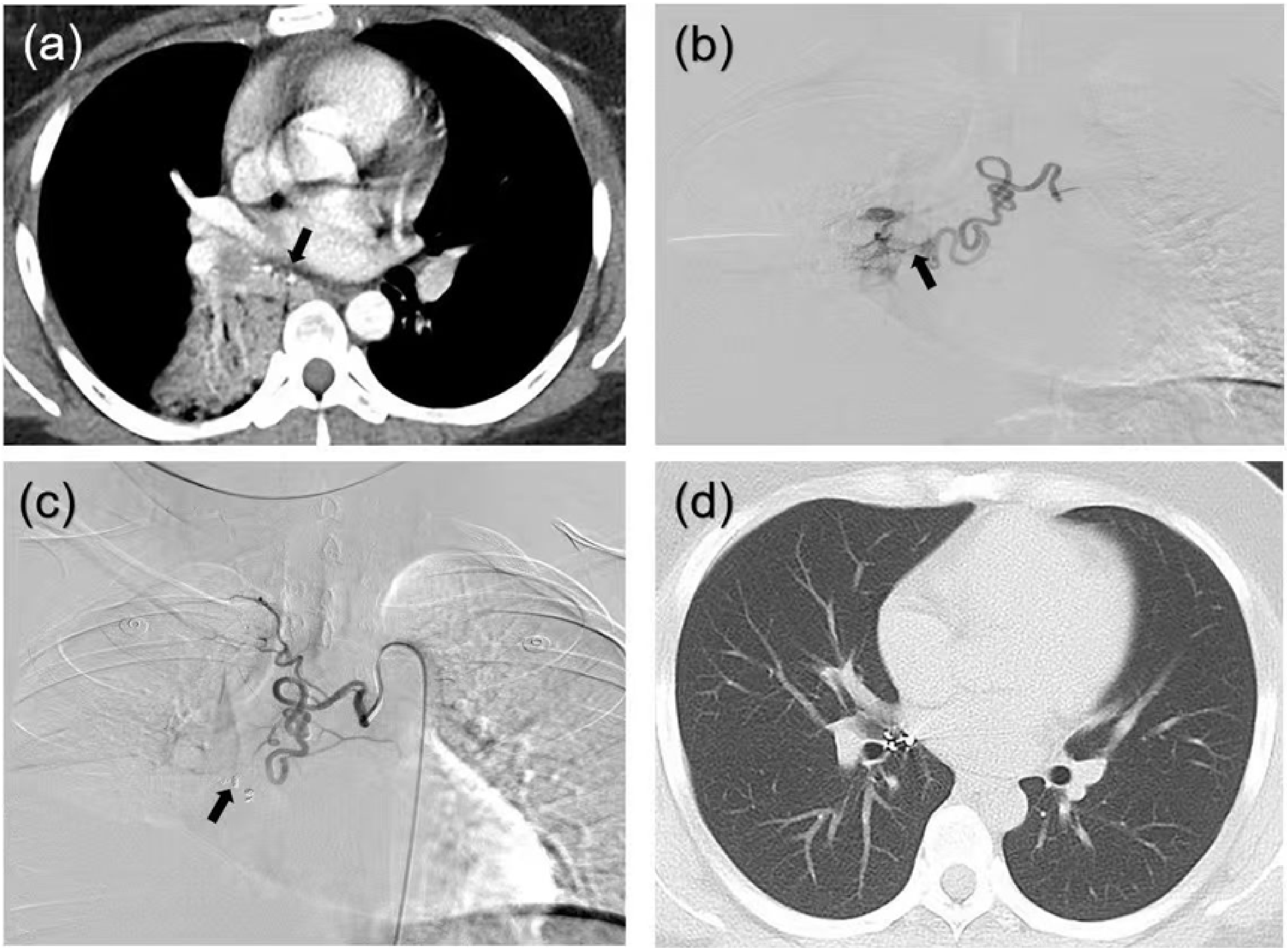

Figure 2: BAE treatment for RHBA. (A) Initial contrast-enhanced thoracic CT: Origin of RHBA from thoracic aorta. Consolidation with bronchial obstruction in right middle and lower lobes. (B) DSA: Tortuous and dilated RHBA with active contrast extravasation. Plexiform vascular network at rupture site. (C) Post-embolization angiography: Successful distal coil embolization of culprit vessel. Complete resolution of abnormal vascular blush. (D) 5-month follow-up non-contrast CT: Complete resolution of bilateral pulmonary patchy opacities. Punctate hyperdensities at right hilum (consistent with embolic material). Satisfactory re-expansion of right middle and lower lobes.

KEYWORDS

Bronchial Artery Racemose Hemangioma (RHBA); Bronchial Artery Embolization (BAE); Interventional radiology; Diagnosis; Embolization strategy

ABBREVIATIONS

RHBA = Primary Bronchial Artery Racemose Hemangioma
BAE = Bronchial Artery Embolization