Received: 10 May 2024 | Accepted: 3 June 2024

DOI: 10.1002/rcr2.1411

CASE SERIES

Respirology Case Reports | WILEY

Bronchial Dieulafoy's disease: A series of seven cases with review of the literature

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Associate Editor: Johnny Chan

Abstract

Bronchial Dieulafoy's disease (BDD), remains poorly understood, with only 88 cases reported globally. Herein, we present the largest case series ($n = 7$) from a single centre, between 2017 and 2023, retrospectively reviewed, detailing clinical presentations, diagnoses, management and up to 4-year follow-up outcomes. Diagnosis relied on characteristic lesions detected through white light bronchoscopy with or without endobronchial ultrasound (EBUS) or narrow band imaging (NBI), along with computed tomography (CT) scans or bronchial angiography. Identification of aberrant vessels beneath lesions and bronchoscopy details were documented. Treatment modalities and follow-up outcomes until December 2023 were noted. All patients were non-smokers. Review of imaging findings by an experienced radiologist was crucial in suspected cases due to risk of bleeding and often unconclusive results from biopsy. Management of BDD varied, with six patients undergoing bronchial artery embolization (BAE) and one requiring lobectomy; four patients received additional endobronchial therapy, one died due to malignancy, none experienced recurrence of haemoptysis. Identifying patients with large volume haemoptysis disproportionate to parenchymal disease in CT scans is important. A bronchoscopic surveillance is crucial to avoid biopsy; it can be confirmed using EBUS of NBI. While no established guidelines exist, BAE and endobronchial therapy emerge as valuable interventions, with surgical resection reserved for recurrent cases.

KEYWORDS
angiography, bronchoscopy, Dieulafoy, haemoptysis, vascular malformations

INTRODUCTION

Dieulafoy's disease is a rare condition characterized by vascular malformation in the submucosa, resulting in the presence of tortuous, dilated vessels. Initially described by Georges Dieulafoy in 1898 as a gastric ulcer, similar lesions have been discovered in the gastrointestinal tract and abdominal organs. Bronchial Dieulafoy's disease (BDD) was first reported in 1995 by Sweerts et al.[1]

Over the past two decades, BDD has been recognized as a cause of massive haemoptysis.[2] Despite the available research, the exact aetiology of BDD remains unknown. Congenital malformation, recurrent mucosal injury due to infection, and tobacco use are among the commonly proposed factors associated with BDD.[3] It is characterized by a small raised non-pulsatile lesion with or without a white cap seen in white light bronchoscopy. Biopsy carries a high risk of life-threatening haemorrhage.[3,4]

Despite the importance of understanding and managing BDD, there have been about 88 reported cases worldwide.[5] Most published data are based only on cases and case series including three or fewer cases. To the best of our knowledge, this is the largest case series of BDD from a single centre in the world to date.[5] In this study, we aimed to describe a series of seven cases managed at an Indian quaternary care centre, focusing on their clinical presentation, diagnosis, management, and follow up. Additionally, we have provided an updated review of the clinical, radiological, and

pathological literature related to this rare but life-threatening condition.

METHODS

Study design and data collection

This retrospective case series was approved by the institutional review board and adheres to the principles outlined in the Declaration of Helsinki. All patients provided written informed consent for the publication of this case series. All patients diagnosed with BDD at our institution over a period of 6 years (2017–2023) were included in this study. Their medical records, chest computed tomography (CT) scans, and bronchoscopy and biopsy reports were retrospectively reviewed, and the data were analysed. All CT images were reviewed by a senior thoracic radiologist, while biopsy slides were examined by a senior thoracic pathologist.

The diagnosis was made based on the following criteria which was defined for this study:

1. Characteristic bronchoscopy appearance of one or more non-pulsatile raised lesions with or without a white cap
   AND
   Narrow band imaging (NBI) showing increased vascularity OR.
   endobronchial ultrasound (EBUS) and Doppler showing a vessel beneath the mucosa.
2. Matching aberrant vessel visualization through either contrast-enhanced chest imaging or bronchial angiography.

We identified seven cases based on the above criteria, which are described below.

Case one

A 15-year-old boy presented with a cough and a 2-month history of small-volume haemoptysis. He was initially evaluated at another centre where they identified a left upper lobe consolidation, while bronchoscopy showed mild bleeding from the left upper lobe and blood clots in the same region. The biopsy showed features of capillary haemangioma leading to his referral to our centre for further management.

During bronchoscopy, clot removal in the left upper lobe's apicoposterior segment revealed a non-pulsatile mucosal bulge prone to bleeding upon contact. A bronchial biopsy, transbronchial lung biopsy, and bronchoalveolar lavage were obtained from the same region, which resulted in significant bleeding controlled by ice cold saline flushes, diluted adrenaline, and electrocautery ablation of the lesion. A biopsy was performed to rule out malignancy.

CT (Figure 1A) showed a prominent bronchial artery in the submucosal region near the origin of the left upper lobe bronchus. Digital subtraction angiogram (Figure 1B) showed tortuous hypertrophied left bronchial arteries and their branches.

The biopsy findings were non-specific and inconclusive. The patient underwent bronchial artery embolization (BAE). However, he suffered a repeat episode of massive haemoptysis a month later, necessitating a second BAE.

Four months later, he had another massive bout and underwent a left upper lobectomy. Figure 2A,B show subbronchial and bronchiolar thick-walled tortuous arteries with occasional fibrin thrombi and the arteries show medial hypertrophy. Notably, the specimen revealed occasional arteries with openings proximal to the bronchial and bronchiolar lumina. The background lung parenchyma showed features of extensive intra-alveolar haemorrhages and mineralization of the alveolar septae.

The patient was followed-up for 4 years post-surgery with no recurrence of symptoms.

Case two

A 59-year-old man, who was a steel plant supervisor and non-smoker, presented with a 2-year history of a cough and streaky haemoptysis, as well as a loss of 15 kg in weight over the past 3 months. His routine evaluation revealed a right lower lobe consolidation. Sputum examination results for tuberculosis were negative.

CT showed a right infrahilar soft tissue density lesion encasing the right lower lobe bronchus with collapse consolidation of the entire right lower lobe. Multifocal consolidatory lesions were identified in both lungs.

Bronchoscopy showed that the segmental bronchi of the right lower lobe were narrowed due to external compression. An enlarged submucosal vessel was noted in the inferior wall of the left upper lobe bronchus suggestive of a Dieulafoy's lesion, confirmed using NBI (Figure 3A). The remainder of the bilateral bronchial trees were normal.

Radial EBUS showed hypoechoic lesions in all segments of the right lower lobe, covering the entire peri-probe area. Cryo transbronchial lung biopsy was performed, along with bronchoalveolar lavage.

A review of the images by a senior thoracic radiologist revealed (Figure 3B) an enlarged submucosal vessel in the left secondary carina at the origin of left upper lobe. A hypertrophied right bronchial artery was also identified.

The biopsy was suggestive of invasive mucinous adenocarcinoma with lepidic and acinar patterns. A positron emission tomography scan revealed bone metastasis. He received palliative radiotherapy for bone metastases, but succumbed to his illness 3 months after diagnosis.

Case three

A 28-year-old lady, a homemaker and non-smoker, with no other comorbidities presented with a 1-month history of a

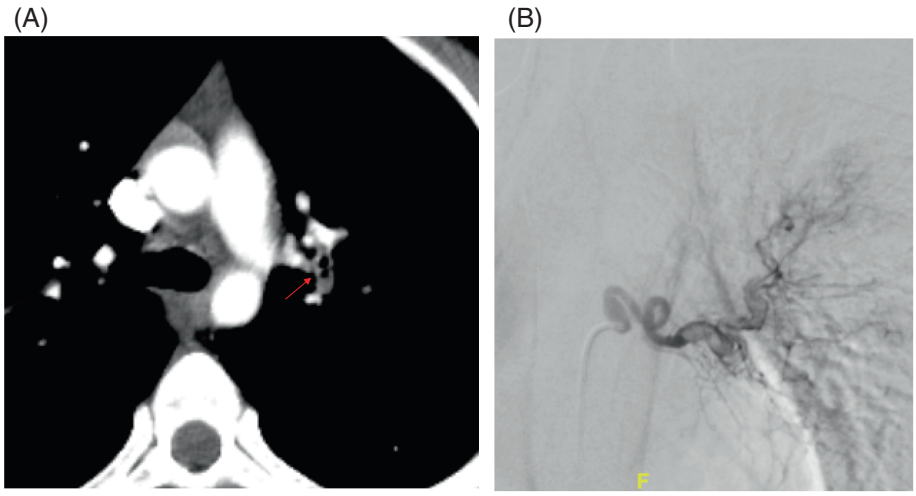

FIGURE 1 (A) Axial sections of CT pulmonary angiogram show a prominent bronchial artery (red arrow) in the submucosal region near the origin of the left upper lobe bronchus. (B) Digital subtraction angiogram shows tortuous hypertrophied left bronchial arteries and their branches. CT, computed tomography.

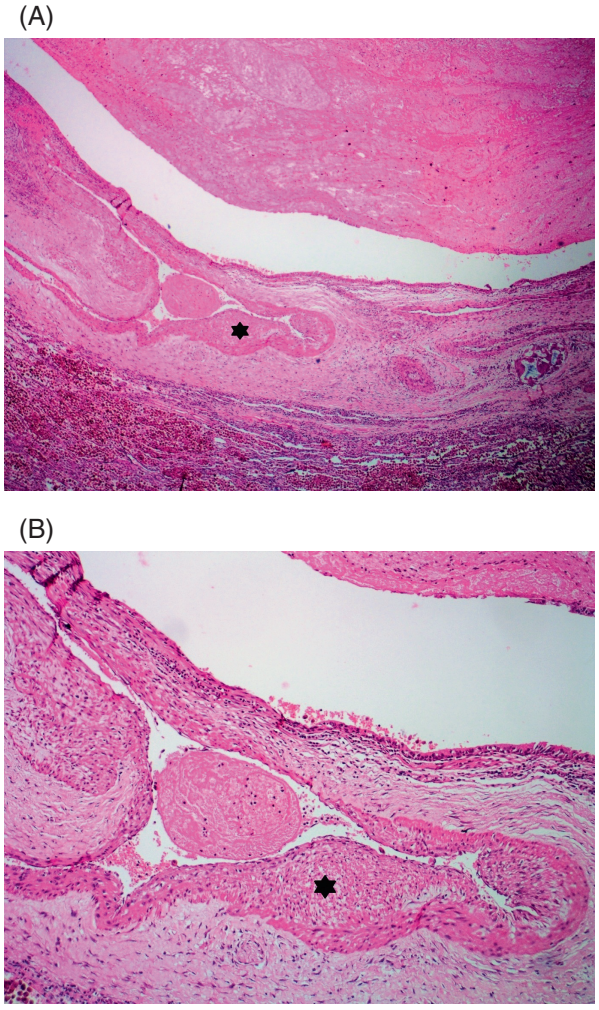

FIGURE 2 (A) Photomicrograph displaying the wall of the bronchus with a dysplastic artery (asterisk) opening close to the lumen, HE, 40×. (B) Photomicrograph displaying the wall of the bronchus with a dysplastic artery (asterisk) opening close to the lumen, HE, 100×.

cough and small-volume haemoptysis. She tested negative for tuberculosis; however, she was treated with anti-tubercular therapy at another centre, which she could not tolerate and hence was referred to our centre for further management.

While a CT pulmonary angiogram ruled out thromboembolism, it incidentally revealed subtle right atrial and ventricular enlargement with apical displacement of the tricuspid valve leaflets and atrialization of the right ventricle, consistent with Ebstein's anomaly. Ill-defined ground glass densities were also found in the posterior segment of the right upper lobe and the superior, anterior basal, and lateral basal segments of the right lower lobe.

Flexible bronchoscopy showed an enlarged mucosal lesion with vascularity beneath the left upper lobe and right lower lobe confirmed by narrow band imaging (Figure 4A). No active bleeding was noted at the time of the bronchoscopy.

A review of the images by a senior thoracic radiologist revealed (Figure 4B) a submucosal bulge due to a hypertrophied left bronchial artery at the origin of the left upper lobe. Small submucosa vessels were noted along the anteromedial aspect of the right lower lobe (Figure 4C).

The patient underwent an elective BAE. Post-embolization bronchoscopy revealed a reduction in size of the right lower lobe lesion, while the lesion in the left upper lobe remained the same. Both were electrocauterized, and there was no residual lesion.

She was kept under follow up for 4 years with no recurrence of haemoptysis.

Case four

A 48-year-old woman, a homemaker and non-smoker, presented with a 2-year history of a cough and small-volume

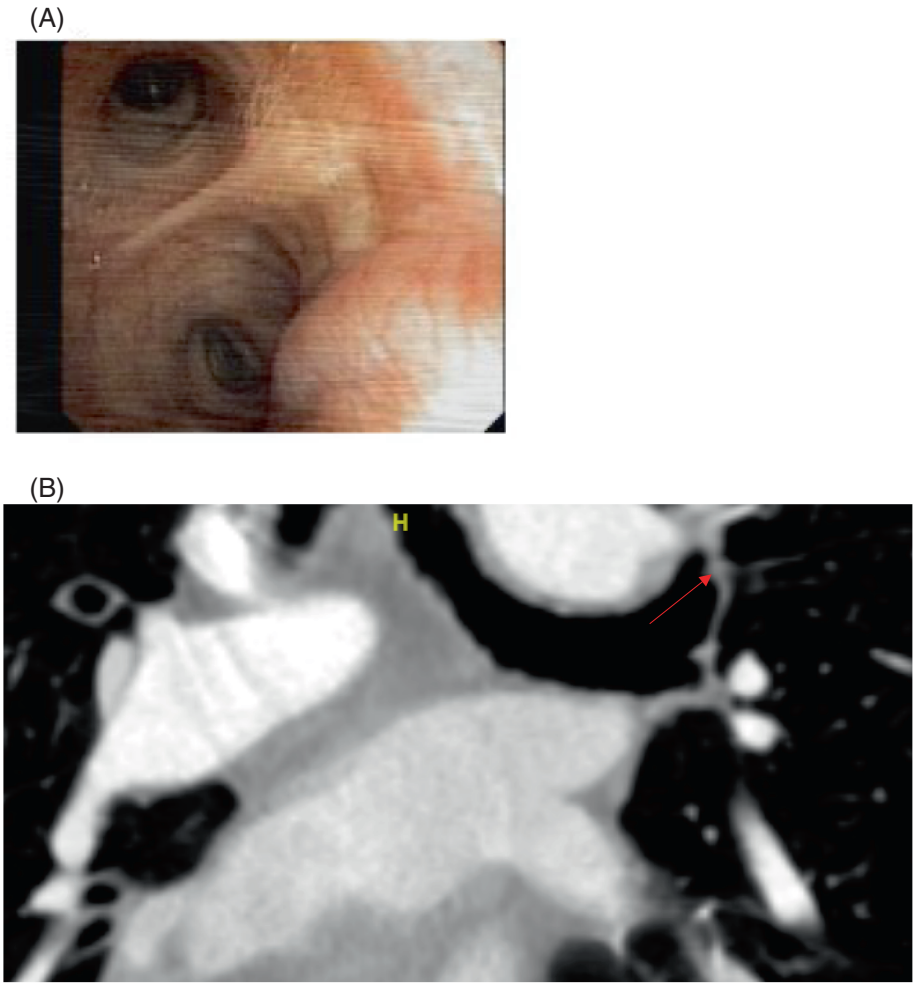

FIGURE 3 (A) An enlarged submucosal vessel in the inferior wall of the left upper lobe bronchus. (B) Coronal sections of CT thorax show an enlarged submucosal vessel (red arrow) in the left secondary carina at the origin of left upper lobe. CT, computed tomography.

haemoptysis. She had previously been treated with multiple courses of antibiotics for the issue.

She had large-volume haemoptysis (50–100 mL), for which she received resuscitation at a local hospital and was referred to our centre for further management.

A CT pulmonary angiogram was performed, which identified an indeterminate right lower lobe nodule.

Flexible bronchoscopy under general anaesthesia revealed a reddish raised lesion in the mucosa of the left main bronchus medial wall, approximately 1.5 cm from the secondary carina (Figure 5A). Profuse bleeding was observed after the first attempt of biopsy, raising the suspicion of Dieulafoy's lesion. Hence, electrocautery ablation was performed at the base of the lesion.

A bronchial angiogram (Figure 5B) showed a hypertrophied bronchial artery supplying both sides, and a careful review of the CT images (Figure 5C) showed the left bronchial artery in close relation to the posterior wall of the left main bronchus, which was embolised.

The patient had no recurrence during the 3-year follow up.

Case five

A 39-year-old man, a truck driver and non-smoker, presented with a 5-year history of a cough and shortness of breath on exertion with occasional streaky haemoptysis. He had two episodes of massive bouts requiring blood transfusion. He was evaluated at a local centre and was diagnosed with pulmonary hypertension based on transthoracic echocardiographic findings.

He visited our centre, and a CT pulmonary angiogram was performed, which showed a chronic pulmonary thromboembolism.

He had an episode of haemoptysis during his hospital stay; hence, an evaluation was planned and bronchoscopy was performed, which revealed blood clots mixed with secretions in the bilateral bronchial tree. The clots were cleared using normal saline instillation and suction. A yellowish mucosal nodule was noted in the left upper lobe side of the left secondary carina. Beneath the nodule, the mucosa bulged and was non pulsatile. A similar, non-pulsatile lesion was found in the left lower lobe. During an attempt to

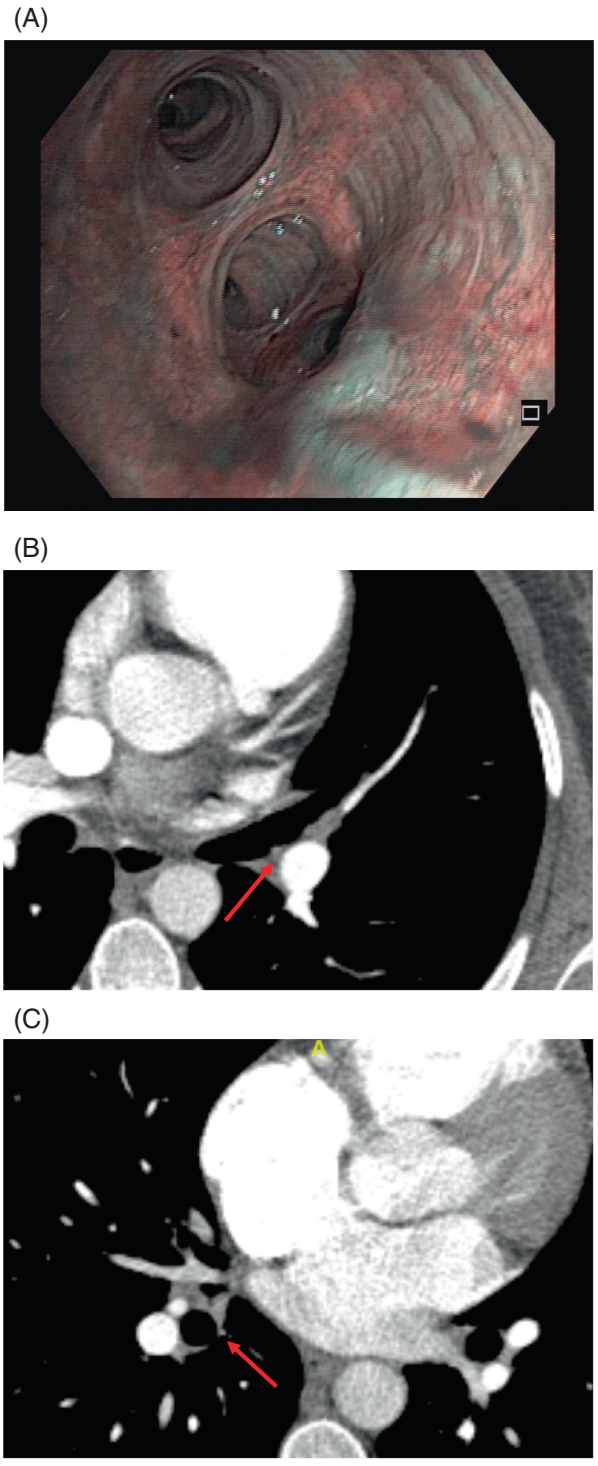

FIGURE 4 (A) Narrow band imaging showing enlarged submucosal vessel at left upper lobe bronchus (B) Axial sections of CT thorax reveal a submucosal bulge (red arrow) due to a hypertrophied left bronchial artery at the origin of the left upper lobe. (C) Axial sections of CT thorax reveal small submucosa vessels (red arrow) along the anteromedial aspect of the right lower lobe. CT, computed tomography.

collect a biopsy sample from the left upper lobe mucosal nodule, moderate bleeding was noted, which was successfully obliterated using a cautery probe. Subsequently an elective BAE was also performed.

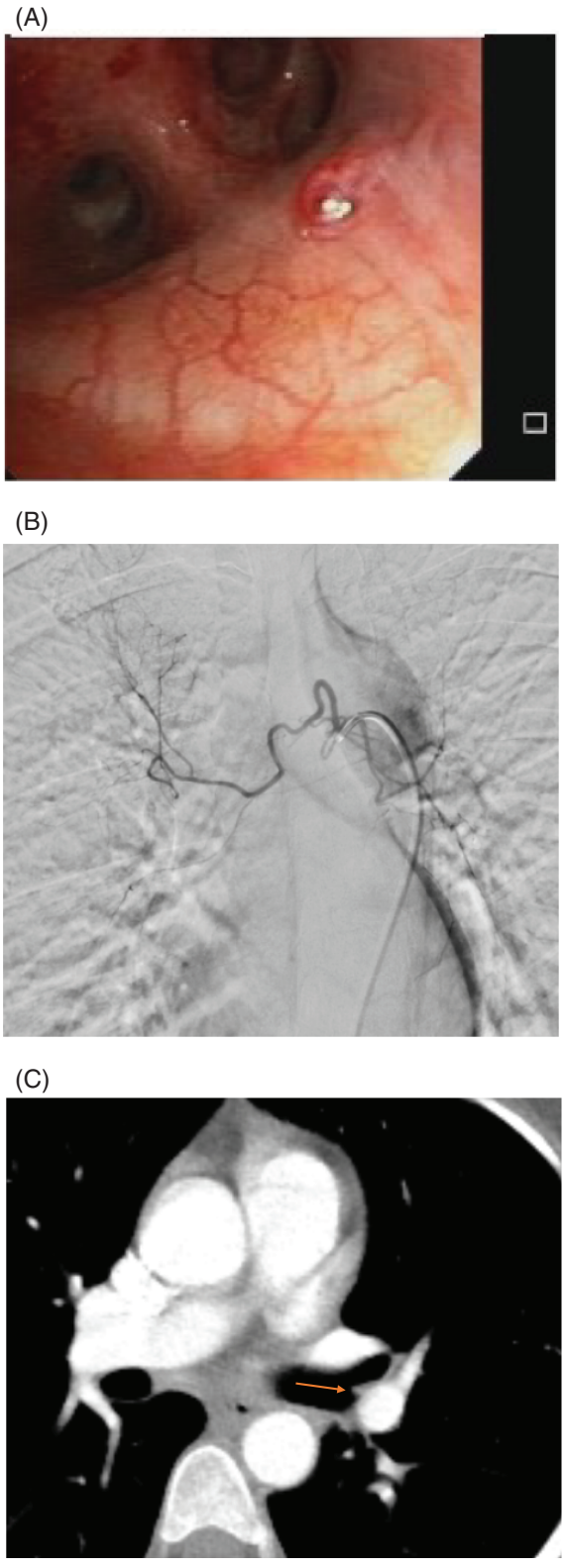

FIGURE 5 (A) A reddish raised lesion in the mucosa of the left main bronchus medial wall, approximately 1.5 cm from the secondary carina in white light bronchoscopy. (B) Digital sub tractional angiogram of bronchial artery shows a hypertrophied bronchial artery supplying both sides. (C) Axial sections of the CT thorax show the left bronchial artery (red arrow) in close relation to the posterior wall of the left main bronchus, which is embolised. CT, computed tomography.

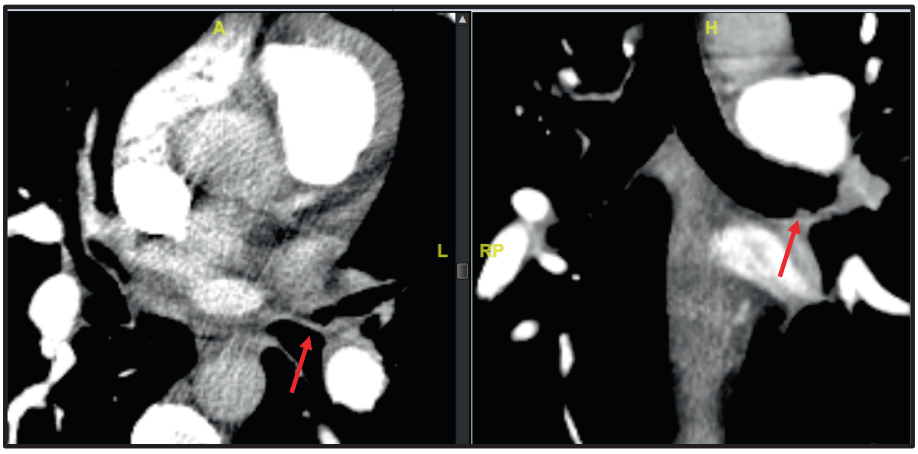

FIGURE 6 Axial sections of the CT thorax and corresponding coronal images reveal a small arterial branch coursing (red arrow) along the submucosal surface of the left upper and lower lobe bronchi. CT, computed tomography.

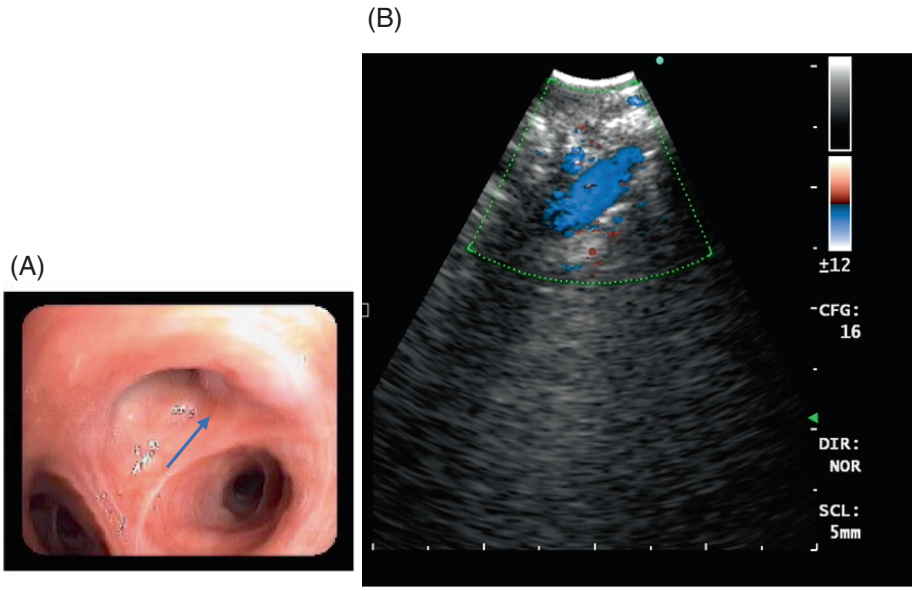

FIGURE 7 (A) White light bronchoscopy image shows a raised non pulsatile mucosal lesion in right upper lobe apical segment (blue arrow). (B) Convex probe EBUS image with Doppler shows the submucosal vessel in the same region. EBUS, endobronchial ultrasound.

The biopsy reports were inconclusive. Correlating CT images (Figure 6) with bronchoscopy findings revealed a small arterial branch coursing along the submucosal surface of the left upper and lower lobe bronchi.

Case six

A 36-year-old woman, a homemaker, with a history of asthma on regular follow-up presented with 2–3 episodes of haemoptysis. Prior imaging results were normal. She was referred to our centre for further evaluation.

Flexible bronchoscopy (Figure 7A) revealed a whitish raised mucosal lesion, which was non-pulsatile at the right upper lobe apical segment. A convex probe EBUS (Figure 7B) revealed a dilatated blood vessel beneath it.

A CT pulmonary angiogram showed a common hypertrophied costo-bronchial trunk. The right bronchial artery (4.5 mm) was seen coursing posterior to the right main bronchus and its branches were seen coursing posterior to the apical segmental bronchus. Furthermore, multilobar mild bronchial and bronchiolar wall thickening present in both lungs were suggestive of airway disease.

She underwent an elective BAE and was followed up for 8 months with no recurrence.

Case seven

A 58-year-old male banker and non-smoker with a known case of idiopathic thrombocytopenic purpura presented with multiple episodes of small-volume and a few episodes of

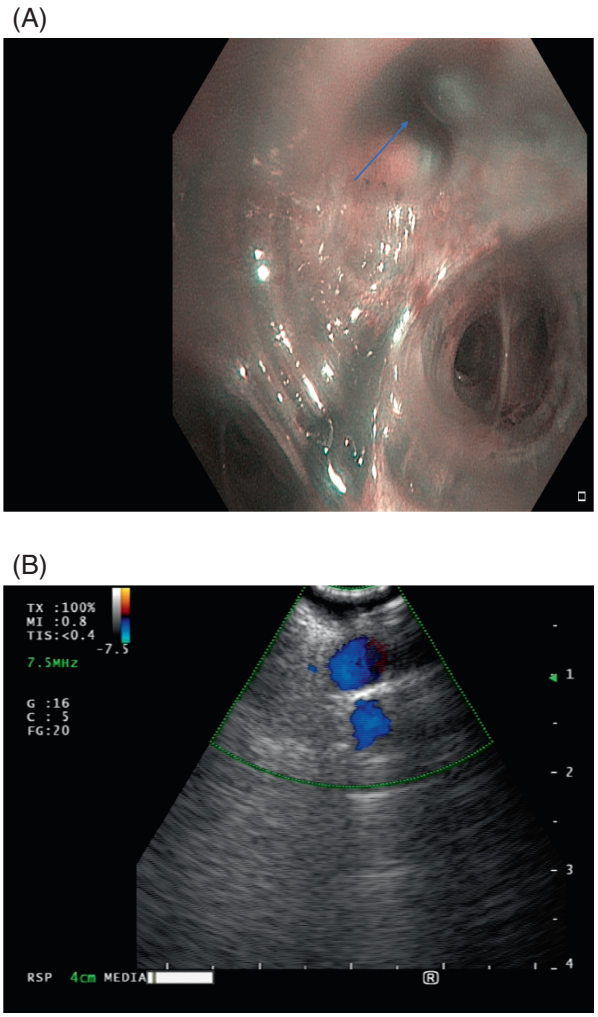

FIGURE 8 (A) Narrow band imaging of the raised vascular lesion at the left upper lobe. (B) Convex probe EBUS image with Doppler shows the presence of a submucosal vessel. EBUS, endobronchial ultrasound.

large-volume haemoptysis. Contrast CT imaging and bronchoscopy findings were normal. The bleeding was attributed to his haematological condition and was referred to our centre for further evaluation.

Flexible white-light bronchoscopy revealed mucosal bulges in the right upper lobe anterior segment and anterolateral wall near the left upper lobe carina, and NBI showed increased vascularity (Figure 8A). Convex probe EBUS identified submucosal vessels within these bulges (Figure 8B). The rest of the bronchial anatomy was normal. Since there was no active bleeding, cautery was not attempted.

A careful review of the images alongside the bronchoscopy report revealed bilaterally hypertrophied bronchial arteries. On the right side, the common costo-bronchial artery was involved, and on the left, the bronchial artery was affected. Notably, a prominent left bronchial artery branch coursed along the submucosal surface of the distal left main bronchus, just proximal to the origin of the left upper lobe bronchus and extending to the left upper lobe.

An elective BAE was performed, and the patient had no recurrence in the last 6 months.

RESULTS

The summaries of the cases are described in Table 1. Among these cases, six patients underwent BAE, whereas one underwent lobectomy after embolization. Three patients received endobronchial cautery or argon plasma coagulation in addition to BAE. Despite controlled bleeding after significant biopsy attempts, the samples showed inconclusive results. The exact pathological diagnosis was only made based on a surgical lobectomy specimen. One patient was found to have an additional lesion in the lung parenchyma, identified as invasive mucinous adenocarcinoma. None of the patients who underwent endobronchial therapy along with BAE experienced a recurrence of haemoptysis during the follow up.

Bronchoscopy

In a suspected case of BDD, a careful surveillance using white light bronchoscopy is crucial as a small raised lesion, which may be non-pulsatile and may or may not have a white cap, can be easily missed.[4] Interestingly, in our series, most cases had lesions on the left side, in contrast to those reported in established literature.[5] To aid in the identification of these lesions, imaging modalities such as NBI can be employed to detect increased vascularity, whereas EBUS with Doppler mode can help visualize the vessels beneath the lesion.[6] It is important to note that performing a biopsy on the lesion can lead to substantial bleeding, and the yield of endobronchial samples is often non-specific. Therefore, biopsy should generally be avoided.[7] If biopsy is attempted, it should be performed in a controlled environment under anaesthesia by an experienced bronchoscopist.[8] In the management of BDD, the use of argon plasma coagulation, in addition to BAE, has shown to be a useful option,[9] we have used electrocautery also as a successful tool for controlling the torrential bleed. These techniques can help control bleeding and provide effective treatment to prevent further bleeding from the affected region.

Radiology

CT findings in BDD can be easily overlooked, potentially leading to unnecessary interventions that may have fatal consequences. The findings on CT scans are often nonspecific, including ground glass opacities, nodules, masses, and abnormal bronchi.[10] Hypertrophied and tortuous bronchial arteries protruding into the lumen can sometimes be identified retrospectively in certain cases, correlating with submucosal tortuous bronchial arteries observed during

TABLE 1 Summary of cases.

	Age (years)/ Sex	Haemoptysis	Concomitant diagnosis	Smoking/ respiratory tract infections	Site of the lesion as per bronchoscopy	Imaging	Treatment	Histopathology	Follow-up
1	15/M	Mild-to-moderate	Nil	Nil	Left upper lobe apicoposterior segment	Prominent bronchial artery along the submucosal region near the origin of the left upper lobe bronchus Digital subtraction angiogram shows tortuous hypertrophied left bronchial arteries and their branches	BAE performed twice followed by LUL lobectomy	Pathology findings[a]	4 years No recurrence
2	59/M	Nil	Lung adenocarcinoma	Nil	Left upper lobe bronchus Radial EBUS showed hypoechoic lesions in all segments of the right lower lobe.	Enlarged submucosal vessel was noted in the left secondary carina, at the origin of the left upper lobe Hypertrophied right bronchial artery Right infrahilar soft tissue density lesion encasing the right lower lobe bronchus with collapse consolidation of the entire right lower lobe Multifocal consolidatory lesions in both lungs	Metastatic adenocarcinoma with poor performance status received best supportive care	EGFR and ALK negative adenocarcinoma—primary lung	Expired 3 months later due to malignancy-related complications
3	28/F	Small-volume haemoptysis	Ebstein's anomaly	Nil	Left upper lobe and right lower lobe	CT—submucosal bulge due to hypertrophied left bronchial artery at the origin of the left upper lobe. Small submucosal vessel noted along the anteromedial aspect of the right lower lobe. Apical displacement of the tricuspid valve leaflets and atrialization of the right ventricle, in keeping with Ebsteins' anomaly. Dilated RA.	BAE followed by bronchoscopy electrocautery	Nil	3-year follow-up No recurrence
4	46/F	Small-volume haemoptysis	Nil	Nil	Left main bronchus lesion noted less than 2 cm from the left secondary carina	One hypertrophied bronchial artery supplying both sides, left bronchial artery seen in close relation to the posterior wall of the left main bronchus.	Bronchoscopic electrocautery followed by BAE	Nil	3-year follow-up No recurrence
5	39/M	Small-volume haemoptysis	Pulmonary embolism and CTEPH	Nil	Left upper lobe side and left lower lobe side of left secondary carina	Chronic pulmonary thromboembolism as evident by occluded and attenuated right upper lobe artery and its branches, as well as the lingular artery and its branches. Eccentric filling defect, cobweb-like filling defects, wall thickening involving left interlobar artery, left lower lobe artery and it branches, scattered subsegmental arterial branches in right middle lobe, right lower lobe, left upper lobe. Small arterial branch is seen coursing along the submucosal surface of left upper lobe and left lower lobe bronchi.	Bronchoscopy electrocautery followed by bronchial artery embolization	Biopsy inconclusive	5-year follow-up No recurrence but worsening CTEPH
6	36/F	Moderate-to-large volume haemoptysis	Nil	Nil	Right upper lobe apical segment	Common hypertrophied Costo bronchial trunk. Right bronchial artery (4.5 mm) is seen coursing posterior to the right main bronchus and its branches are seen coursing posterior to the apical segmental bronchus. Multilobar mild bronchial and bronchiolar wall thickening present in both lungs.	BAE	Nil	8-month follow-up No recurrence

TABLE 1 (Continued)

	Age (years)/ Sex	Haemoptysis	Concomitant diagnosis	Smoking/ respiratory tract infections	Site of the lesion as per bronchoscopy	Imaging	Treatment	Histopathology	Follow-up
7	57/M	Moderate-volume haemoptysis	ITP	Nil	Right upper lobe anterior segment Left main bronchus–anterolateral wall near left upper lobe	Hypertrophied bronchial arteries (right-common costo-bronchial, left bronchial artery) bilaterally. Prominent left bronchial artery branch coursing along the submucosal aspect of distal left main bronchus, just proximal to the origin of left upper lobe bronchus and the right upper lobe	BAE	Nil	1-month follow-up No recurrence

Abbreviations: ALK, anaplastic lymphoma kinase; BAE, bronchial artery embolization; CT, computed tomography; CTEPH, chronic thromboembolic pulmonary hypertension; EBUS, endobronchial ultrasound; EGFR, epidermal growth factor receptor; ITP, idiopathic thrombocytopenic purpura; LUL, left upper lobe; RA, rheumatoid arthritis.

[a]Arteries opening close to the bronchial and bronchiolar lumen. The background lung parenchyma shows features of extensive intra-alveolar haemorrhage and mineralization of the alveolar septae.

bronchoscopy.[11] Ground glass opacities related to alveolar haemorrhage are often present on CT scans. The presence of clots within the bronchial lumen can indicate the site of bleeding. While bronchial Dieulafoy's lesions are typically found in the right bronchus, this case series identified lesions in the left main bronchus or at the left secondary carinal division.[12]

Pathology

BDD is characterized by a dysplastic artery, exhibiting a tortuous appearance, within the submucosa of the bronchus. Before diagnosing BDD, it is crucial to exclude chronic lung diseases such as bronchiectasis, which can present with thick-walled ectopic bronchial arteries.[13] However, in small biopsy specimens, the tissue may often be non-diagnostic, displaying normal mucosa or being contaminated by bleeding. The pathogenesis of this disease remains uncertain, with the origin of the anomaly considered congenital, acquired, or a variation of normal. Commonly affected sites include the oesophagus, jejunum, duodenum, gall bladder, colon, and rectum.[13]

DISCUSSION

The primary clinical presentation of BDD is haemoptysis, which can vary in volume from small to large, with most cases showing normal findings on high-resolution CT imaging.[14] Bronchoscopic examination results typically reveal small protrusions in the bronchial mucosa covered by a white cap. Utilizing techniques such as EBUS and NBI can help visualize the underlying vascular lesions. It is important to note that if these lesions are identified, biopsy should be avoided due to the potential for difficult-to-control and life-threatening bleeding.[15]

The aetiology of BDD in our case series is mainly related to congenital malformation as most patients were non-smokers which is in contrast to a proposed aetiology, moreover most patients had associated etiologies like malignancy, Ebstein anomaly, chronic thromboembolism or immune thrombocytopenia hence we propose this may be a part of systemic dysregulation and aberrant vascularity.[16]

Currently, no established guidelines exist for managing this rare disease. Endobronchial therapy using electrocautery or argon plasma coagulation is an option for experienced interventional pulmonologists. Bronchial arterial angiography and embolization should be attempted in all cases, as the source of bleeding is systemic in more than 80% of cases.[17] Surgical resection of the affected lobe is considered the treatment of choice in recurrent cases.[18,19]

Diagnosing BDD requires a high index of suspicion in patients presenting with moderate to massive haemoptysis and normal CT findings without structural lung

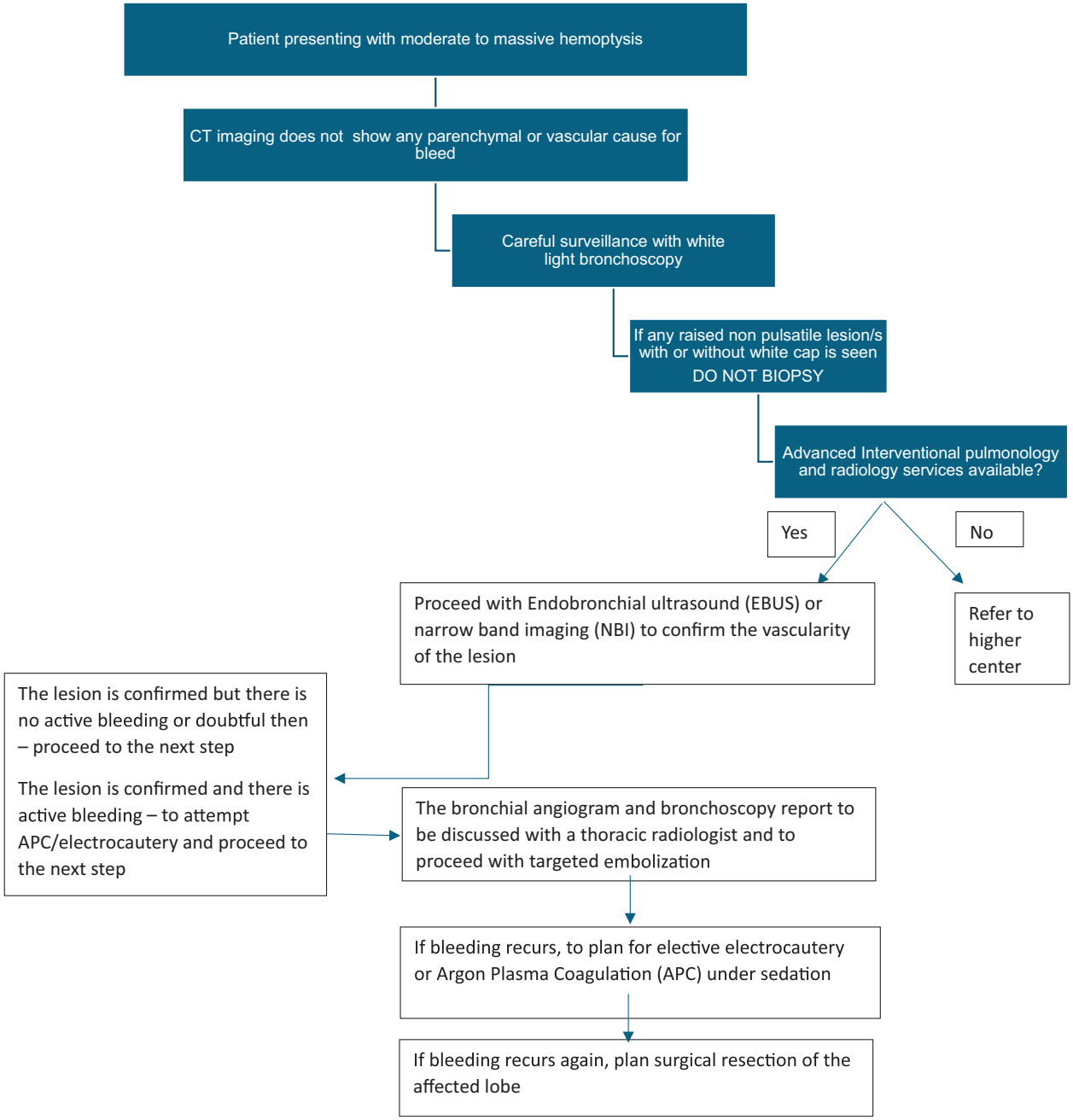

FIGURE 9 Algorithm for a suspected case of Bronchial Dieulafoy's disease.

damage (Figure 9). Systematic bronchoscopic surveillance is crucial for identifying the characteristic lesion, which can be confirmed using NBI and EBUS. Bronchial angiography should be performed to detect aberrant vessels, and embolization should be considered.[20] Biopsy of the lesion can lead to torrential bleeding, making it crucial to have the necessary facilities to manage such cases. In instances of active bleeding, local electrocauterization or argon plasma coagulation, along with BAE, should be attempted.[21] Surgical resection should be considered in recurrent cases and is considered the gold standard treatment.[20–22]

ACKNOWLEDGMENTS

The authors acknowledge the contributions of Mr Karunakaran and Mr Naresh—respiratory therapists for bronchoscopy reports and images. Mrs Nadhiya office secretary for patient communication.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images. This retrospective case series was approved by the institutional review board.

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How to cite this article: Nair AA, James P, Vimala LR, Kodiatte T, Gupta R. Bronchial Dieulafoy's disease: A series of seven cases with review of the literature. Respirology Case Reports. 2024;12(6):e01411. https://doi.org/10.1002/rcr2.1411