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CASE REPORT

**Nurda ŞİMŞEK
VESKE¹(ID)
Ekrem Cengiz
SEYHAN¹(ID)
Demet TURAN¹(ID)
Neslihan FENER²(ID)**

- ¹ Department of Chest Disease, University of Health Sciences Yedikule Chest Diseases and Thoracic Surgery Training and Research Hospital, İstanbul, Türkiye
² Department of Pathology, University of Health Sciences Yedikule Chest Diseases and Thoracic Surgery Training and Research Hospital, İstanbul, Türkiye

Endobronchial vascular lesions of the lung

ABSTRACT

Endobronchial vascular lesions of the lung

Primary vascular lesions of the lung are rare and are usually described in isolated case reports or small series. The lesions may be intraparenchymal, but they can also be seen endobronchially, most commonly in the trachea and rarely in the distal airways. This case series highlights the need for careful evaluation of endobronchial vascular lesions, which may present with mass-like imaging findings or have a polypoid appearance and pose a risk of major bleeding during biopsy.

Key words: Capillary hemangioma; arteriovenous hemangioma; endobronchial lesion; hemoptysis; arteriovenous malformation

ÖZ

Akciğerin endobronşial vasküler lezyonları

Akciğerin primer vasküler lezyonları nadirdir ve genellikle izole vaka çalışmaları veya küçük seriler olarak bildirilmiştir. Bu lezyonlar intraparankimal yerleşimli olabileceği gibi daha çok trakeada ve nadiren de olsa distal havayollarında endobronşial olarak da gözlemlenebilir. Bu olgu serisi, kitle benzeri görüntüleme bulguları veya polipoid görünümde olabilen ve biyopsi sırasında majör kanama riski oluşturabilen endobronşiyal vasküler lezyonların dikkatli değerlendirilmesinin gerekliliğini vurgulamaktadır.

Anahtar kelimeler: Kapiller hemanjiom; arteriovenöz hemanjiom; endobronşial lezyon; hemoptizi; arteriovenöz malformasyon

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Address for Correspondence

Dr. Nurda ŞİMŞEK VESKE
Department of Chest Disease, University of Health Sciences Yedikule Chest Diseases and Thoracic Surgery Training and Research Hospital, İSTANBUL-TÜRKİYE
e-mail: nrdnsimsek@gmail.com

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

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INTRODUCTION

Endobronchial vascular lesions of the lung are rare. In addition to surgical treatments, pedunculated lesions can be treated endobronchially via rigid bronchoscopy. We aimed to compile three separate cases with endobronchial components, which are rarely observed in the literature, and highlight the unique features of each.

Case Presentation

Patient 1

A 48-year-old female patient with a history of hypertension and rheumatoid arthritis was referred to our clinic after mediastinal lymphadenopathy had been detected via thoracic computed tomography (CT) performed during an evaluation for a breast mass

(Figure 1A). The patient had no remarkable habits or family history, and her hemogram and biochemical parameters were normal. A lesion was incidentally detected in the trachea during endobronchial ultrasonography (EBUS) to evaluate the mediastinal lymph nodes. A pedunculated, polypoid lesion was present on the posterior wall approximately 6-7 cm from the vocal cords (Figure 2A). Rigid bronchoscopy

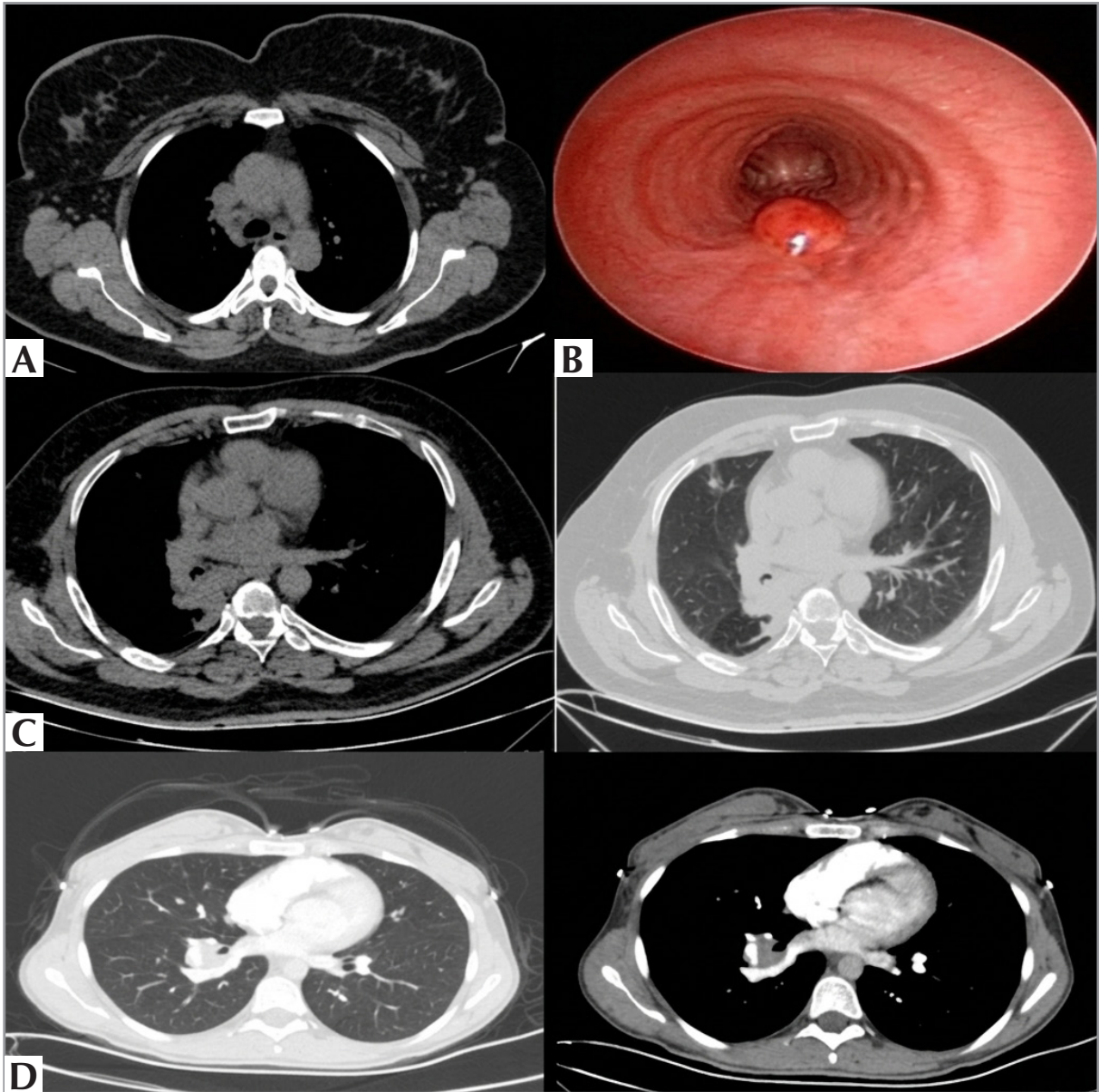


Figure 1. Patient 1's thoracic CT scan of an axial section, the mediastinal window shows an image of a normal tracheal air column (A). Patient 1's FOB image (B). Patient 2's thoracic CT image of the axial, mediastinal and parenchymal windows showing a mass lesion protruding into the bronchus in the lower lobe (C). Patient 3's thoracic CT of axial section, mediastinal and parenchymal windows showing soft tissue density lesion in the lower lobe (D).

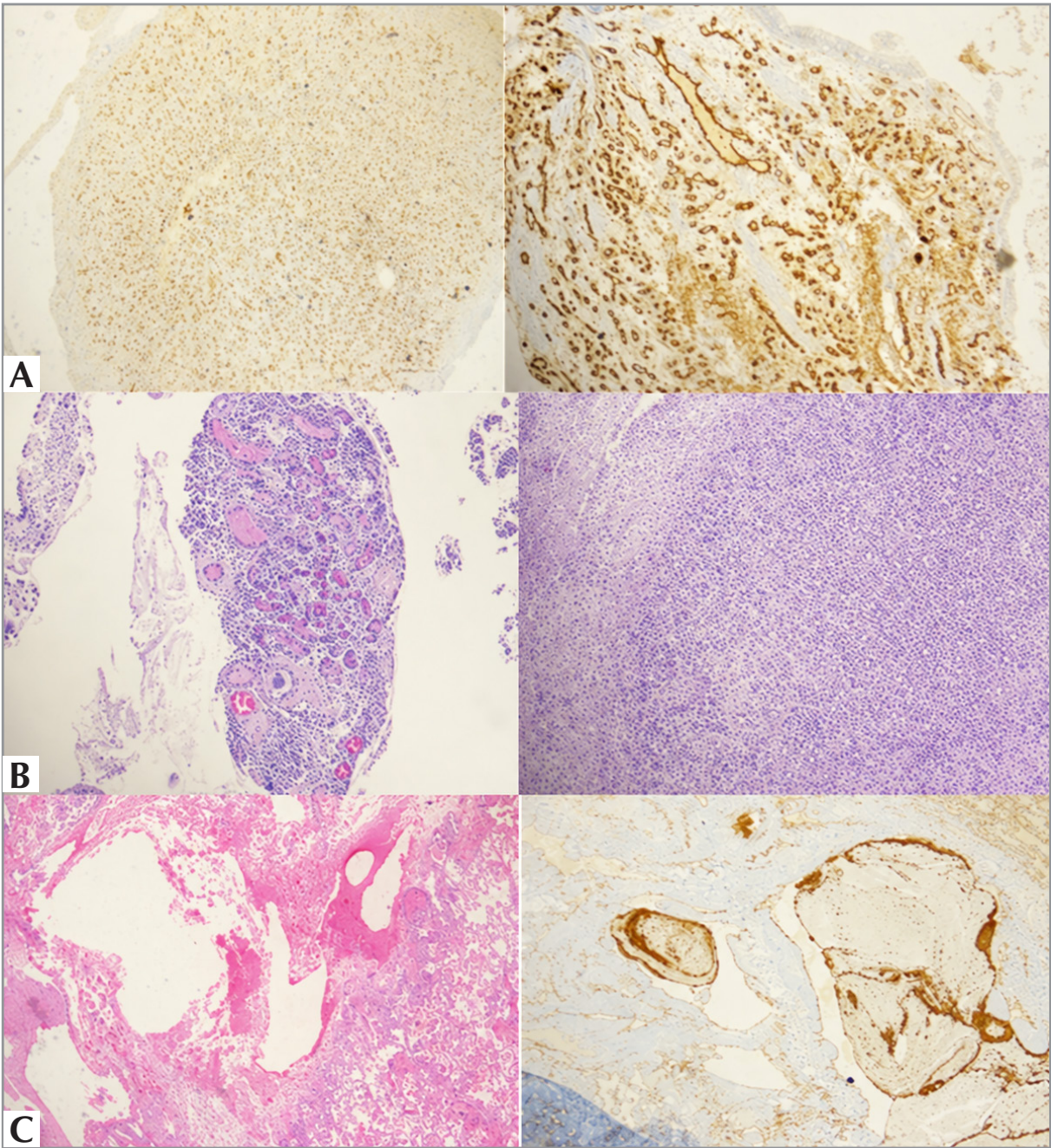


Figure 2. (20X10 H&E stain) Proliferating capillary vessel structures and capillary hemangioma. Positive reaction with CD31 (A). (20X10 H&E stain) Capillary proliferation in the bronchial wall, hemangioma. Positive reaction with CD31 (B). (20X10 H&E stain) Thick-walled anastomosed vascular structures, positive reaction with CD31 (C).

was planned to protect the airway in case of possible bleeding and manage bleeding more safely due to the location and appearance of the existing lesion, and the lesion was not intervened during the EBUS procedure. The lesion was removed via a snare during rigid bronchoscopy, and argon plasma coagulation (APC)

was applied to the residual tissue. The procedure was completed without complications. Histopathological examination revealed a capillary hemangioma (Figure 1C). No recurrence was observed during follow-up fiberoptic bronchoscopy (FOB) at six months.

Patient 2

A 63-year-old male patient presented with a cough and bloody sputum. The patient, a foreign national, had been diagnosed with lung cancer in his home country and had a history of chemotherapy and radiotherapy. No information was obtained regarding the treatment applied or the type of tumor other than the use of pazopanib. There were no features in his family history or habits. The hemogram, biochemistry, and laboratory tests regarding bleeding diathesis were normal. On thorax CT, there were lesions protruding from the right lower lobe bronchus into the lumen (Figure 1C). A vegetative lesion was observed, almost completely obstructing the right lower lobe of the patient who underwent FOB due to hemoptysis. Bleeding that developed after biopsies was controlled with local adrenaline and cold saline. No massive bleeding was observed. Biopsies revealed an arteriovenous hemangioma (Figure 2B).

Patient 3

A 24-year-old female presented with intermittent hemoptysis for one month. She had no significant personal or family history, and her hemogram, biochemical values, and coagulation studies were normal. Thoracic CT revealed a soft tissue density at the entrance of the lower lobe (Figure 1D). Bronchoscopy revealed increased vascularity with a bulging lesion at the middle lobe entrance and a clot obstructing the lower lobe entrance. While attempting to clear the clot, profuse bleeding occurred from the basal segments, which could not be controlled. The patient underwent emergency thoracotomy, and a right bilobectomy was performed. Pathology revealed arteriovenous-lymphatic cavernous and capillary combined vascular malformations, widespread parenchymal hemorrhage, cavernous hemangioma, and arteriovenous malformation (Figure 2C). These findings extended from the surgical margin distally, involving the entire bronchial mucosa in full thickness and partially infiltrating the surrounding parenchyma. The postoperative follow-up was uneventful.

DISCUSSION

Primary vascular neoplasms of the lung are rare and are typically described in isolated case reports or small series. Due to their rarity, clinical and radiological findings are often mistaken for more common

conditions until a histopathological diagnosis is established. The most common vascular-origin tumors in the pulmonary system include benign lesions such as hemangiomas and lymphangiomas, as well as malignant tumors such as epithelioid hemangioendotheliomas, angiosarcomas, and Kaposi sarcomas. Non-neoplastic vascular abnormalities, including arteriovenous malformations and arteriovenous hemangiomas, may also appear as mass-like lesions on imaging.

These lesions are typically located within the lung parenchyma but may also occur endobronchially, most commonly in the trachea and, less frequently, in distal airways.

Endobronchial hemangiomas should be considered in the differential diagnosis of endobronchial lesions. Hemangiomas are common benign tumors of the head and neck region in childhood, often with multiple localizations that regress spontaneously during adolescence. They are extremely rare in adults (1). Most of the cases reported in the lung are located in the trachea, and a case of isolated capillary hemangioma detected as endobronchial has also been reported.

Since tracheal capillary hemangiomas are small, chest X-rays and thoracic CT results are usually normal (2). The bronchoscopic appearance is non-specific and may mimic adenoid cystic carcinoma or carcinoid tumors (2). Treatment includes Neodymium-doped Yttrium Aluminum Garnet (Nd:YAG) lasers, APC, angiographic embolization, endotracheal brachytherapy, and excisional forceps biopsy. Patients generally respond well to these methods, with favorable outcomes, and are monitored for recurrence. Patients may present with hemoptysis and cough, but, as in our first case, they may also be asymptomatic. A pedunculated polypoid lesion was incidentally detected during a bronchoscopy performed for mediastinal lymph node transbronchial needle aspiration under EBUS guidance to evaluate mediastinal lymph nodes detected on thoracic CT performed to assess malignancy metastasis. An endobronchial treatment approach with rigid bronchoscopy was applied, and no recurrence was observed during follow-up. In these cases, both surgical and endobronchial approaches have been described in the literature, as well as serial imaging methods for asymptomatic patients (3,4).

Primary pulmonary hemangiomas are rare pulmonary neoplasms, and cavernous variants are extremely rare in the lungs (3). These tumors may involve the bronchi or lung parenchyma, be single or multiple, and can occur in syndromes such as Osler–Weber–Rendu syndrome and Blue rubber bleb nevus syndrome (3,5). Clinical presentation may range from asymptomatic to chronic cough, worsening dyspnea, and recurrent or even massive hemoptysis (4). Both surgical and endobronchial treatment approaches can be applied. Cavernous hemangiomas (CHs) are generally found in the skin, subcutaneous tissues, muscles, and liver. Pulmonary cavernous hemangiomas (PCHs) are extremely rare and typically present with non-specific clinical symptoms.

PCHs frequently occur at various points in the lung parenchyma and lower respiratory tract but do not extend into the bronchial lumen. PCHs can occur at any age, and although some sources mention a slight female predominance and a frequency of localization in the right lung, there is no definitive data. The frequency of occurrence in different lung lobes is generally accepted to be equal (6). Our third patient, as in the present data, was a young female patient and the lesion was located in the right lung. However, the difference in our patient was that the lesion extended into the bronchial lumen.

Cases of CHs associated with adenocarcinomas of the gastrointestinal system and lung have been reported (7).

Some endobronchial vascular lesions, such as arteriovenous malformations, can appear mass-like and cause massive bleeding during biopsy, as observed in our third case. Therefore, when pulsatile, tubular, and hypervascular lesions are identified, the equipment and environment for managing potential massive bleeding should be evaluated before any intervention.

CONCLUSION

Notably, vascular lesions may have different appearances, and lesions with a tumor-like appearance and increased vascular components may bleed massively during biopsy. Angiographic imaging protocols should be preferred when vascular pathology is suspected. We would like to emphasize that endobronchial treatment methods are promising both as a diagnostic and therapeutic method for pedunculated

lesions, that the association of adenocarcinoma and cavernous hemangioma can be observed in the lung as in gastrointestinal system malignancies, and that the absence of uptake on positron emission tomography imaging should alert the physician to the possibility of vascular pathology.

PATIENT CONSENT

Patient consent was obtained.

Peer-review: Externally peer-reviewed.

CONFLICT of INTEREST

The authors have no conflict of interest to declare.

AUTHORSHIP CONTRIBUTIONS

Concept/Design: NŞV, ECS, DT

Analysis/Interpretation: NŞV, ECS

Data acquisition: NŞV, ECS

Writing: DT, ECS, NŞV

Clinical Revision: ECS, NF

Final Approval: ECS, NF

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