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Case report

Hemomediastinum due to spontaneous rupture of a mediastinal bronchial artery aneurysm – A rare cause of thoracic pain

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A B S T R A C T

Hemomediastinum is a rare pathological event. Multiple underlying causes and contributory factors can be identified, such as trauma, malignancy, iatrogenic, bleeding disorder or mediastinal organ hemorrhage. Also, a mediastinal bronchial artery aneurysm may be the source of a hemomediastinum. Hemoptysis is an important directive symptom, however occasionally, patients only present with thoracic pain or symptoms related to extrinsic compression of the airways or esophagus. Using contrast-enhanced computed tomography (CT) of the chest, hemomediastinum can be adequately diagnosed, and the involved vascular structures can be revealed. In case of a (ruptured) bronchial artery aneurysm, transcatheter embolization provides a minimally invasive procedure and is treatment of first choice. In this case report, a 76-year-old female is presented with spontaneous rupture of a mediastinal bronchial artery aneurysm resulting in hemomediastinum causing thoracic pain. Superselective embolization of the left bronchial artery was successfully performed.

Introduction

Spontaneous hemomediastinum is rarely observed in clinical practice and is a potentially life-threatening condition. Underlying causes have been categorized into three groups. First, spontaneous hemomediastinum may occur secondary to bleeding disorders such as hemophilia, or secondary to anticoagulant treatment. Secondly, mediastinal tumors (e.g. thymomas, teratomas), organs or blood vessels may be involved. Thirdly, one can distinguish spontaneous idiopathic hemomediastinum, which can particularly appear after sudden increase in intrathoracic pressure, (e.g. during coughing, sneezing or vomiting, or sudden sustained hypertension) [1,2]. In case of a primary problem of (large) blood vessels, the most common cause is aortic (aneurysm) dissection. Rupturing of a mediastinal bronchial artery aneurysm is a rather unusual cause of spontaneous hemomediastinum. Bronchial artery aneurysms are detected in less than 1% of all patients who undergo selective bronchial angiography [5].

Case

A 76-year-old female patient with past medical history including acute rheumatic fever (childhood), osteoporosis and total abdominal hysterectomy presented herself at the emergency room because of acute thoracic pain. Apart from the pain, which radiated to both jaws and upper back, the patient had no other complaints. She was not using any medication. Physical examination was normal, with a blood pressure of 155/75 mmHg (equal in right and left arm), a regular pulse of 70 beats per minute and a temperature of 36.3 °C. The electrocardiography was normal, and no abnormalities were observed upon chest radiography and echocardiogram. Laboratory blood testing showed normal kidney and liver function, normal coagulation, a normal blood count and negative cardiac enzymes. The patient was discharged from the hospital with a diagnosis of atypical thoracic pain.

Two weeks later she attended the emergency room again with similar severe thoracic pain. She also mentioned complaints of heart burn and dysphagia. A contrast-enhanced chest CT was performed upon suspicion of a pulmonary embolism. The CT

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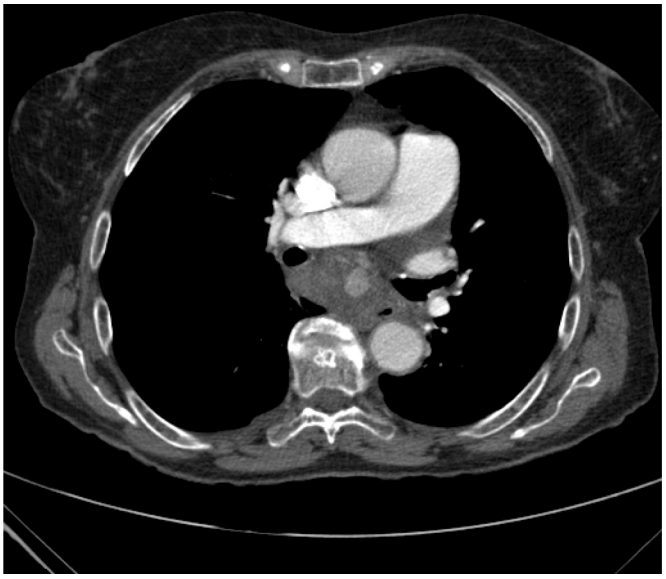

Fig. 1. Contrast-enhanced CT of the mediastinal level shows a big mass with contrast extravasation indicating active bleeding in the tumor.

ruled out a pulmonary embolism, but did reveal a large mass in the posterior mediastinum (Fig. 1) with an axial diameter of 5.5 cm and cranio-caudal diameter of 7.4 cm, that extended to the subcarinal level (Fig. 2). This mass showed contrast extravasation suggesting active bleeding. Angiography was performed, demonstrating a large (pseudo)aneurysm of the left bronchial artery (Fig. 3). Superselective embolization using coils was successfully carried out (Fig. 4) in a coaxial way, using a 5F Cobra catheter and a microcatheter for superselective embolization. Fibered coils were placed distally and proximally to the pseudoaneurysm in order to avoid backflow and consecutive recurrence. The patient recovered quickly and was discharged two days after the procedure. Six weeks later, upon evaluation at the outpatient clinic, she was free of complaints and chest CT showed that the mediastinal hematoma had completely resolved (Fig. 5).

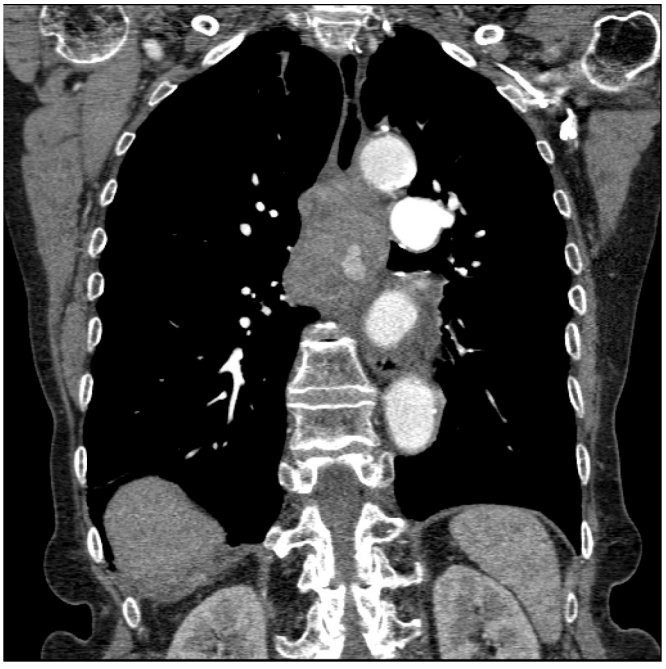

Fig. 2. Coronal multiplanar reformations show the cranio-caudal extend of the mass.

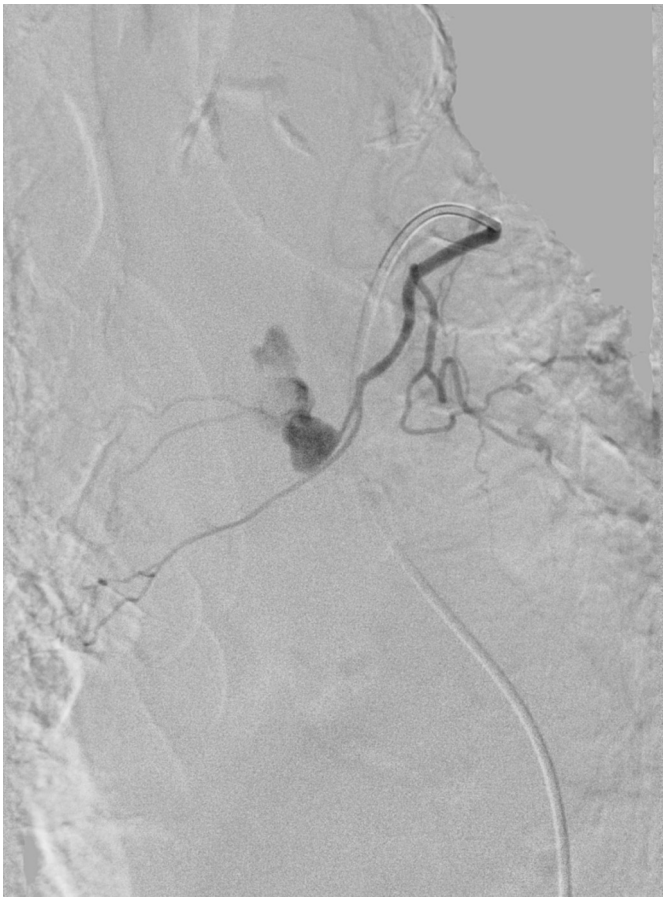

Fig. 3. Selective angiography shows a large (pseudo)aneurysm of the left bronchial artery.

Discussion

Aneurysms and pseudoaneurysms of the pulmonary vasculature are rare and more often affect the pulmonary arteries than the bronchial arteries or the pulmonary veins [5]. An aneurysm typically involves all 3 layers of the vessel wall, whereas a pseudoaneurysm represents a contained rupture in which not all layers of the affected wall are involved. Bronchial arteries are normally $<$1.5 mm in diameter at their origin and decrease to 0.5 mm as they enter the broncho-pulmonary segment. A bronchial artery diameter exceeding 2 mm is generally considered pathological and associated with an increased risk of severe clinical complications [3].

Bronchial artery aneurysms may be mediastinal or intrapulmonary in location and are associated with different medical conditions: congenital (sequestration, pulmonary agenesis), arteriovenous malformation, vasculitis (Behçet disease, Hughes-Stovin syndrome), bronchiectasis, infectious disease (tuberculosis, atypical mycobacteria, aspergillosis, histoplasmosis), sarcoidosis, silicosis, post-traumatic, hereditary hemorrhagic telangiectasis (Osler–Weber–Rendu disease) or idiopathic [5]. In many of the before-mentioned diseases, pulmonary circulation is reduced at the level of the pulmonary arterioles because of hypoxic

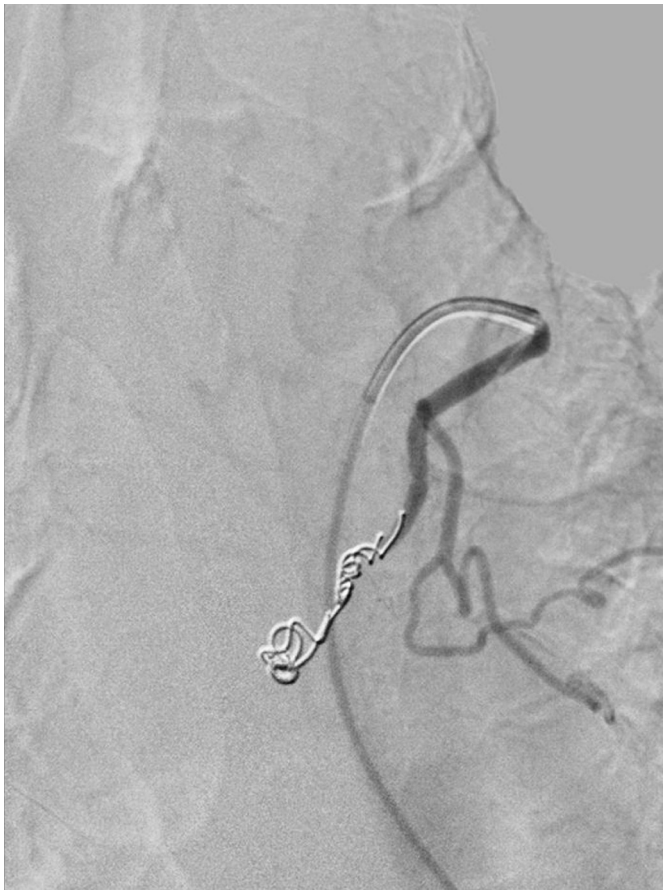

Fig. 4. Superselective coil embolization was performed with full occlusion of the feeding artery using fiber coils.

vasoconstriction, thrombosis and vasculitis inducing a compensatory enlargement of the bronchial arteries [4].

The clinical presentation of a bronchial artery aneurysm depends on its size and location, but also on the presence of concomitant disease. Intrapulmonary bronchial artery aneurysm is commonly manifested by hemoptysis which can range from blood-streaking of sputum to massive hemoptysis that is potentially life-threatening. Patients with a (ruptured) mediastinal bronchial artery aneurysm more frequently present with chest pain and with symptoms related to extrinsic compression of adjacent structures such as the airways (shortness of breath), the esophagus (dysphagia) or the vena cava (vena cava superior syndrome) [1,2,5,6]. Sporadically, a hemothorax is found.

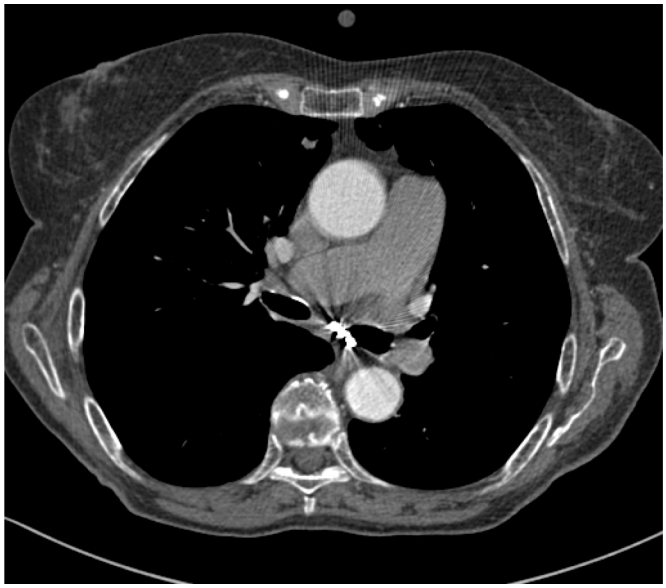

Fig. 5. CT follow-up demonstrates full occlusion of the feeding artery and no active bleeding with disappearance of the mediastinal hematoma.

In order to adequately diagnose a hemomediastinum, performing a chest CT with contrast material application is the designated approach. Consecutive angiography may then be the next best step towards treatment.

Obviously, a ruptured bronchial artery aneurysm requires immediate treatment, but also an asymptomatic bronchial artery aneurysm should generally be treated, as rupture can be dangerous. Surgical extirpation can be done through (video-assisted) thoracotomy and reliably eliminates the lesion, but is invasive and not feasible in every patient. In our opinion, transcatheter embolization is the treatment of first choice. Superselective catheterization with a microcatheter is usually performed using coils for safe embolization. Careful evaluation of potential spinal cord branches should be carried out prior to embolization to avoid severe complications (e.g. spinal cord ischemia, which is extremely unlikely to happen with this type of embolization). Furthermore coil embolization should be performed by placing coils proximal and distal from the pseudoaneurysm in order to avoid recurrence. After the procedure, patients may experience chest pain (prevalence 24–91%) or dysphagia (prevalence 0.7–18%); both are likely related to an ischemic event caused by embolization and are usually transient. Subintimal dissection of the bronchial artery can occur (prevalence 1–6.3%), but is usually asymptomatic. High success rates have been reported for bronchial artery embolization, but recurrence after successful embolization can occur—probably due to collateral vessels, incomplete embolization, and arterial re-canalization—making re-intervention necessary [1,4,6].

Conclusion

A hemomediastinum is a rare pathological event with several possible underlying causes including a ruptured bronchial artery aneurysm. Bronchial artery aneurysms present with various symptoms ranging from massive hemoptysis to subtle chest pain. First choice treatment consists of transcatheter embolization.

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