Recurrent massive hemoptysis due to postbronchotomy bronchial artery aneurysm: A case report

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Bronchial artery aneurysm (BAA) is a rare clinical entity. A case of intrapulmonary BAA associated with previous bronchotomy at the same site is described. A 22-year-old woman, who had undergone bronchotomy of the intermediate bronchus for the removal of a foreign body four years previously, presented with recurrent hemoptysis. Because of an increased risk for spinal cord ischemia, she immediately underwent lung resection rather than therapeutic embolization. The incidental finding of a BAA of any cause cannot be assumed to be stable, and immediate management should be undertaken regardless of the presence or absence of symptoms.

Key Words: *Bronchial artery aneurysm; Massive hemoptysis; Surgical management*

Bronchial artery aneurysm (BAA) is a rare clinical entity, which is detected in fewer than 1% of all patients selected for bronchial arteriography (1). In some patients, BAA is complicated by rupture or hemoptysis, both of which may be life-threatening. In the majority of cases, the location of aneurysmal dilation is at or near the bronchial artery origin from the thoracic aorta. We report a case of intrapulmonary BAA located adjacent to the intermediate bronchus, associated with previous bronchotomy and recurrent episodes of hemoptysis. The mechanism of formation, clinical presentation and management are discussed.

CASE PRESENTATION

A 22-year-old woman presented to the emergency department with recurrent massive hemoptysis and mild dyspnea. On admission, the patient was conscious and tachypneic (20 breaths/min). Her blood pressure was normal (120/80 mmHg), and her heart rate was slightly raised (100 beats/min). Her serum hemoglobin level was 98 g/L.

Four years previously, she had been subjected to bronchotomy, at the level of the intermediate bronchus by right posterolateral thoracotomy, for the removal of a foreign body due to persisting pneumonia of the right lower lobe.

An enhanced computed tomography (CT) scan revealed a small mass adjacent to the intermediate bronchus that was

Une hémoptysie massive récurrente causée par un anévrisme de l'artère bronchique après une bronchotomie : Un rapport de cas

L'anévrisme de l'artère bronchique (AAB) est une entité clinique rare. Est décrit un cas d'AAB intrapulmonaire associé à une bronchotomie antérieure, au même foyer. Une femme de 22 ans, qui avait subi une bronchotomie de la bronche intermédiaire pour extraire un corps étranger quatre ans auparavant, s'est présentée en raison d'une hémoptysie récurrente. Compte tenu d'un risque accru d'ischémie médullaire, elle a immédiatement subi une résection pulmonaire plutôt qu'une embolisation thérapeutique. Il ne faut pas penser qu'un AAB de quelque cause découvert par hasard est stable, et il faut prévoir une prise en charge immédiate, en présence ou en l'absence de symptômes.

suspicious for BAA. A bronchoscopy confirmed the origin of hemoptysis at the level of the intermediate bronchus. Because the patient was hemodynamically stable, she was a possible candidate for therapeutic intervention by BAA embolization. The patient was immediately transferred to the angiography suite and subjected to elective bronchial angiography. An aneurysm of the median bronchial artery (size 3 mm) was depicted adjacent to the intermediate bronchus (Figure 1). To exclude the BAA from the circulation, the median bronchial artery, along with the fifth to eighth intercostal arteries, would need to be embolized.

Because of an increased risk for spinal cord ischemia, it was decided to proceed immediately to surgery rather than to perform embolization. A right posterolateral thoracotomy through the fifth intercostal space was performed. Because of the previous operation at the same site, BAA dissection was difficult. The median bronchial artery was ligated.

Postoperative recovery was uneventful. At a follow-up four months later, her condition remained good.

DISCUSSION

BAA is a rare condition (2-8). It is classified as mediastinal, intrapulmonary, or both. In most cases, the aneurysm is located near the origin of the bronchial artery (1-8). Our patient had a BAA at the level of the intermediate bronchus, which is an

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Figure 1 Preoperative selective bronchial artery angiography depicting a bronchial artery aneurysm

even more unusual condition. Furthermore, in our patient, the cause of BAA was trauma from previous bronchotomy at that location. To our knowledge, this is the first time that a post-bronchotomy BAA has been reported.

BAAs may occur as isolated lesions or as associations with mediastinal or intrapulmonary lesions. They are usually detected in cases of bronchiectasis, or in cases of recurrent or persistent bronchopulmonary inflammation. Other causes believed to predispose patients to BAA development are atherosclerosis (3), infection, trauma (9) and Rendu-Osler-Weber syndrome.

In most of the cases, BAA was incidentally identified on chest CT during investigations of bronchiectasis or other bronchopulmonary disease (3,4). They usually appear as a suspicious mediastinal or intrapulmonary mass.

BAA can remain asymptomatic for a long period before rupture takes place. The condition may present as hemoptysis (2,4), hematemesis (3), hemothorax (6) or mediastinal hemorrhage (1). When rupture occurs into the pulmonary parenchyma, hemoptysis is the main clinical sign (1,2,4,9). If the rupture takes place into the mediastinum or the pleural space, the clinical presentation may simulate that of aortic dissection or rupture (1,6).

There are insufficient clues about the process of aneurysm rupture, and the size of the BAA seems to be irrelevant to that complication. In general, increased bronchial arterial flow to the lungs due to various factors is the suggested mechanism of bronchial artery dilation in most cases. In our case,

the etiological process of BAA development was previous surgical trauma. Arterial wall injury led to decreased arterial wall strength, bronchial artery dilation and finally to aneurysm formation. BAA may erode or rupture into the adjacent bronchial tree due to pressure necrosis.

Although contrast-enhanced chest CT discloses a high index of suspicion for intrapulmonary BAA (1,2), the variety of structures or possible anatomical anomalies in the area make differential diagnosis extremely difficult. The interpretation of chest CT scan cannot lead to a solid diagnosis. Definitive diagnosis of the cause of hemoptysis was confirmed by selective bronchial artery angiography. In most cases, selective bronchial arteriography also has a therapeutic role. Embolization of the bronchial artery, as well as the associated intercostal arteries, is usually the first-line management of BAA on either an urgent or elective basis (1). A detailed understanding of bronchial anatomy is required to avoid potential complications, such as transverse myelitis (2,10).

Therefore, the recommended treatment for stable patients is embolization of the suspicious vessels during selective bronchial arteriography, followed by anatomical resection of the affected lung parenchyma in cases of recurrent hemoptysis (11).

Mediastinal or intrapulmonary BAA, manifested with hemoptysis and associated with severe ipsilateral bronchiectatic lesions, should be managed with lobectomy (4), especially in the cases of massive hemoptysis. The rationale for lung resection is that by removing the etiological factor of BAA development, the patient is no longer predisposed to the formation of a new one in the future. Moreover, sole embolization may not be possible to solve the problem of hemoptysis. In the present case, BAA was speculated to be the cause of hemoptysis. However, other sites of the bronchiectatic lobe may be the source of intra-bronchial bleeding. Moreover, the destroyed pulmonary parenchyma should also be removed. Femorofemoral bypass is recommended in cases of uncontrolled hemorrhage due to BAA rupture into the mediastinum or pleural space. In one case (12), the successful treatment of major hemoptysis in cystic fibrosis with tranexamic acid, in an individual in whom 12 previous bronchial artery embolization procedures had been performed and further procedures were contraindicated secondary to bronchial artery to spinal artery collaterals, was described.

The incidental finding of a BAA of any cause cannot be assumed to be stable. Taking into consideration the published case reports of spontaneously ruptured BAA that led to life-threatening hemorrhage, immediate management should be undertaken regardless of the presence or absence of symptoms.

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