Open Surgical Correction of Multiple Bronchial Artery Aneurysm: A Case Report

Jef Van den Eynde[1], Arian Arjomandi Rad[2], Ilya Karasov[3], Arkady Airapetyan[3], Michel Sá[4], Jerry Easo[5], Konstantin Zhigalov[6], Alexander Weymann[6], and Bakytbek Kadyraliev[7]

[1]KU Leuven University Hospitals Leuven
[2]Imperial College London
[3]Perm State Medical Academy named after E A Vagner
[4]Pronto Socorro Cardiológico of Pernambuco
[5]Heart Center Essen-Huttrop gGmbH
[6]University Hospital Essen
[7]S.G. Sukhanov Federal Center of Cardiovascular Surgery, E.A. Vagner Perm State Medical University

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Abstract

Bronchial artery aneurysms are a rare vascular entity. They can have various presentations ranging from an incidental finding on radiological examination to life-threatening hemoptysis. We report the case of a 60-year old woman with three posterior mediastinal bronchial artery aneurysms who presented with unilateral periscapular pain, shortness of breath, hoarseness, and dysphagia. The bronchial artery aneurysms were removed successfully via thoracotomy, with excellent recovery and relief of the periscapular pain. We use this case as a platform to discuss unilateral periscapular pain as an atypical referral pattern for a bronchial artery aneurysm, as well as implications for treatment.

INTRODUCTION

Bronchial Artery Aneurysm (BAA) is an extremely rare presentation consisting of enlargement of bronchial arteries with potentially fatal hemorrhagic consequences [1]. The first reported case of bronchial aneurysm goes back to 1930 in a patient with syphilis [2, 3]. Incidence in patients undergoing selective bronchial angiography is estimated as fewer than 1% [4]. Classification of these aneurysms is based on their location, being either mediastinal or intrapulmonary [5]. While the first presentation might result from an incidental finding on radiological examination, the rupture of the aneurysm can lead to a mediastinal hemorrhage, being potentially fatal for the patient. Moreover, the condition can often mimic acute aortic syndromes (such as aortic dissection) and is often associated with pulmonary agenesis, chronic lung inflammation, bronchiectasis, and vascular pathology [6,7]. The literature reports several cases of endovascular treatment for BAA with either transcatheter arterial embolization or aortic stent graft [8], but surgical options are also available. We hereby present the case of three BAA treated through thoracotomy approach.

CASE REPORT

A 60-year-old woman was admitted to the hospital due to right periscapular dull pain, which worsened with changes in body position. She also complained from shortness of breath, hoarseness, and dysphagia. The patient had no known history of atherosclerosis, chronic or acute lung disease, or thoracic trauma. A chest X-ray revealed the presence of an expanded shadow at the right lung hilum with clearly defined oval

structures. Multispiral computed tomography (MSCT) (Figure 1) and selective angiography of the right bronchial artery (Figure 2) were performed, revealing 3 bronchial artery aneurysms (BAA) with cavities, measuring 76mm, 25mm, and 22mm. Aneurysms were located in the posterior right mediastinum under the tracheal bifurcation.

The patient was referred for surgery. A left-side thoracotomy was performed along 4 intercostal spaces, after which the pulmonary ligament, esophagus, left main bronchus, and tracheal bifurcation were mobilized. Consequently, the descending aorta and its branches were isolated and access to the contralateral pleural cavity was obtained. In the posterior mediastinum, three aneurysms were found: one large aneurysm measuring 7cm (Figure 3), and two smaller aneurysms measuring 1.5cm and 2cm, respectively. The feeding arteries were identified and stapled, followed by excision of the aneurysms. The incision was closed and drains were positioned. The patient was transferred to the intensive care unit (ICU) with no complications.

Mechanical ventilation took 10 hours, and the patient could be discharged from the ICU after 48 hours. After the drains had been removed, the patient was transferred to the general ward. Postoperative course was uneventful. According to post-operative control echocardiography and MSCT, the patient showed no abnormalities and her initial symptoms had been relieved. The patient was discharged 13 days after surgery.

DISCUSSION

BAA is a rare vascular entity which can be broadly classified based on its localization into either mediastinal or intrapulmonary BAA [4]. Although all BAA can be found incidentally in patients with no symptoms, the latter frequently presents with hemoptysis. Mediastinal BAA on the other hand can present with a variety of symptoms resulting from extrinsic compression of contiguous structures including atelectasis, dysphagia, and superior vena cava syndrome [9]. Occasionally, the aneurysm may rupture, resulting in an acute and life-threatening condition characterized by chest pain, hemothorax, hemomediastinum, hematemesis, and shock [10].

In our case, the presenting symptom was unilateral periscapular pain, which is an atypical referring pattern for pain originating from the mediastinum. Although such a pattern has been demonstrated before in thymic carcinomas [11], lymphomas [12], or sarcomas [13] infiltrating the intercostal nerves, the pericardium, or vertebrae, such infiltration was not revealed on imaging in our patient. The viscerosomatic convergence theory might therefore provide a better explanation [14]. This theory states that noxious stimuli from a diseased organ can transmit to an adjacent normal structure, resulting in functional changes in the latter. This phenomenon is mediated by convergence of visceral and somatic afferent neurons at the level of the spinal cord, in lamina I and V of the dorsal horn, and results in afferent stimuli of the viscera being interpreted by the brain as dermatomal and sclerotomal pain. Viscerosomatic convergence might therefore be a possible mechanism of pain in patients presenting with BAA. In addition to the periscapular pain, our patient experienced shortness of breath, hoarseness, and dysphagia, all symptoms that are compatible with extrinsic compression of surrounding structures by the BAA.

In conclusion, we presented the case of a 60-year old woman with unilateral periscapular pain as an atypical presentation of three posterior mediastinal BAAs. The diagnosis was suspected based on chest X-ray and consequently confirmed on chest CT and selective bronchial arteriography. BAAs were removed successfully via thoracotomy, with excellent recovery and relief of the periscapular pain.

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FIGURE LEGENDS

Figure 1. **(A, B)** CT-reconstruction of right pleural cavity and mediastinum. **(C, D)** CT scans. Arrows show a large aneurysm of the right bronchial artery measuring 76 mm in diameter.

Figure 2. Angiography of the right bronchial artery.**(A)** Bronchial artery. **(C1-3)** Cavities of the aneurysms.

Figure 3. Large aneurysm after surgical excision.