Case report

Intralobar lung sequestration with systemic coronary arterial supply

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Abstract. Pulmonary sequestration is a rare anomaly. An accurate pre-operative evaluation of its vascular supply is essential for the surgeon’s operative approach. We describe here an intrapulmonary sequestration with vascular arterial supply via the left circumflex and the right coronary artery. This case demonstrates that if aortography is unrevealing, then a coronary source should be considered in the preoperative search for the arterial supply to a pulmonary sequestration. Moreover, pulmonary sequestration should be listed in the differential diagnosis of aberrant coronary arteries.

Key words: Intralobar lung sequestration – Bronchiectases – Right coronary artery

Introduction

Pulmonary sequestration, diagnosed in the adult, is a rare anomaly. Because of several possible complications, pulmonary sequestration must be resected [1, 2]. For the surgeon’s tactical approach it is essential to know the exact vascular supply of the sequestration, as indicated by several reports of operation-associated fatal hemorrhages [2]. Pulmonary sequestrations often are supplied by one or more arteries originating from the descending aorta, or by one of the aortic side branches. There are cases in which retrograde aortography failed to demonstrate the vascular supply of a pulmonary sequestration [2, 3]. We now report the case of a patient with a pulmonary sequestration receiving its arterial supply via two sources, the left circumflex and the right coronary artery. Arterial feeding of a pulmonary sequestration via a single coronary artery has been described twice [1, 4].
nary sequestration. The lung emphysema was pronounced on the left side, possibly secondary to the chronic inflammatory process. The presence of an atelectasis of the whole left lower lobe, which could have led to a compensatory overinflation, was ruled out by CT and chest X-ray.

Thoracic aortography excluded an additional accessory arterial supply. The findings were compatible with an intralobar lung sequestration possessing a main arterial supply from the right coronary artery; however, a malignant process could not be ruled out.

At operation, an intralobar sequestration of the lung was confirmed, and in the surrounding lung areas severe inflammatory changes were observed. The left lower lobe of the lung had to be resected, and in the lingular segment bullous, degenerated regions also had to be removed by wedge resection. The pathologic specimen showed bronchiectases behind proximal obliterated bronchi. There was severe, chronically active, purulent bronchitis and severe chronic pneumonia characterized by interstitial fibrosis and dense-packed arterial and venous lung vessels sometimes resembling an arteriovenous aneurysm. There was marked intimal fibrosis of the pulmonary arteries and chronic lymphadenitis of the regional lymphatic nodes.

After the operation, the patient was well enough to be discharged from the hospital after a few days and felt considerably better.

Discussion

The patient presented with typical clinical signs of an intralobar lung sequestration. The onset of symptoms of chronic obstructive pulmonary disease was present already in childhood, and the symptoms were stable for years. The correct diagnosis was finally made by chance when the patient presented with chest pain and was admitted for angiographic evaluation.

In neonates or young children, chest radiographs demonstrating abnormal lung or vascular shadows suggest the diagnosis of pulmonary sequestration [5]; in adults the differential diagnosis is much broader. In the literature there are only two recent reports of pulmonary sequestrations receiving arterial blood supply via a coronary artery [1, 4], whereas the origin of bronchial arteries from the coronary vasculature was previously known [6].

In this patient the presence of a malignant process had to be considered because there were enlarged lymph nodes and the onset of B symptoms such as loss of weight. The definite diagnosis could only be reached by resection of the process. Even in patients with an intralobar sequestration without any symptoms surgery should be considered because there is always the possibility of a microscopic infection causing later occurrence of symptoms [1]; moreover, there are reports of fatal pulmonary hemorrhages secondary to intralobar sequestration [2].

Traditionally, the definitive diagnosis of pulmonary sequestration is established by an arteriogram demon-