

Sequestered Hypoplastic Pulmonary Lobe Supplied by the Circumflex Coronary Artery in a Patient with Coronary Artery Disease: A Case Report

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ABSTRACT

Bronchopulmonary sequestrations are usually supplied by single or multiple branches from the descending aorta. We report on a rare form of sequestration of a hypoplastic mid-pulmonary lobe with arterial supply from the circumflex coronary artery coincidentally discovered during coronary angiography in a female patient with coronary artery disease and a history of recurrent hemoptysis. The patient underwent myocardial revascularization and resection of the sequestered lobe in a single-stage approach.

CASE REPORT

A 64-year-old woman with unstable angina, previous percutaneous transluminal coronary angioplasty, and stent implantation to the anterior descending coronary artery was admitted to our hospital for cardiac catheterization. The examination revealed a complete obstruction of the previously stented artery (left anterior descending artery [LAD]) as well as a 90% stenosis of the first diagonal artery. The right coronary artery was a wide vessel collateralizing with the LAD. The ejection fraction was normal. Contrast medium injection to the ostium of the left main coronary artery revealed the presence of an atrial branch arising from the proximal part of the circumflex artery. Several terminal branches arising from this aberrant artery were feeding a vascular bed located in the mid-right pulmonary field. (Figure 1A). Although chest radiographs were normal, computed tomography showed an abnormal mid-lobe mass with multiple cystic elements. Contrast enhanced magnetic resonance angiography revealed the presence of a well-delimited consolidation in the right lung with aberrant blood supply, confirming the diagnosis of intralobar sequestration.

The patient did not mention any episode of pulmonary infection, but a more detailed history revealed 3 episodes of modest hemoptysis accompanied by chest discomfort. In view of these findings, the patient was referred for surgical revascularization and resection of the intralobar sequestration.

A median sternotomy was performed, followed by wide opening of the pericardium and the right pleural space. Exploration of the mediastinum did not reveal any visible morphological abnormality. Soon after the right lung was freed from adhesions, a hypoplastic, nonventilated mid-lobe with multicystic appearance was detected (Figure 2). This small and nonfunctional lobe had clear landmarks although fused with the upper pulmonary lobe.

Hilar dissection did not reveal any pulmonary artery branch supplying this abnormal lobe. The only vessel found during dissection was a short affluent vein draining into the right lower pulmonary vein. After ligation and division of the affluent vessel, the lobe was divided from the upper lobe using a stapling instrument. Several small arteries surrounding the mid-lobe bronchus were divided by cautery, and the bronchus was stapled. Exploration of the divided bronchus revealed a narrow, nearly nonexistent lumen. After a lobectomy, cardiopulmonary bypass was established and myocardial revascularization was performed. The left internal mammary artery was anastomosed to the LAD and a saphenous vein graft was anastomosed to the first diagonal branch. The postoperative course was uneventful and the patient was discharged on the tenth postoperative day. A cardiac catheterization was performed on the ninth postoperative day to assess the results of the surgical treatment. The examination confirmed the disappearance of the sequestration feeding vessels (Figure 1B) and the patency of the grafts.

DISCUSSION

Intralobar sequestrations receiving arterial supply from the coronary circulation are rare entities with few cases reported in the literature. Even more rare are sequestrations originating from the circumflex and, to our knowledge, there are 5 previous reports. Silverman and associates reported the first case of left lung sequestration supplied by the circumflex coronary artery, found in a 66-year-old man with angina and recurrent pulmonary infections [Silverman 1994]. The patient underwent myocardial revascularization without resection of the sequestration to avoid mediastinal contamination. Temes reported a case of mid-right lung sequestration supplied by an aberrant artery arising from the circumflex artery in a 51-year-old man with anxiety disorders [Temes 1998]. Venous drainage was to the left atrium. Coronary arteries were normal and the patient was

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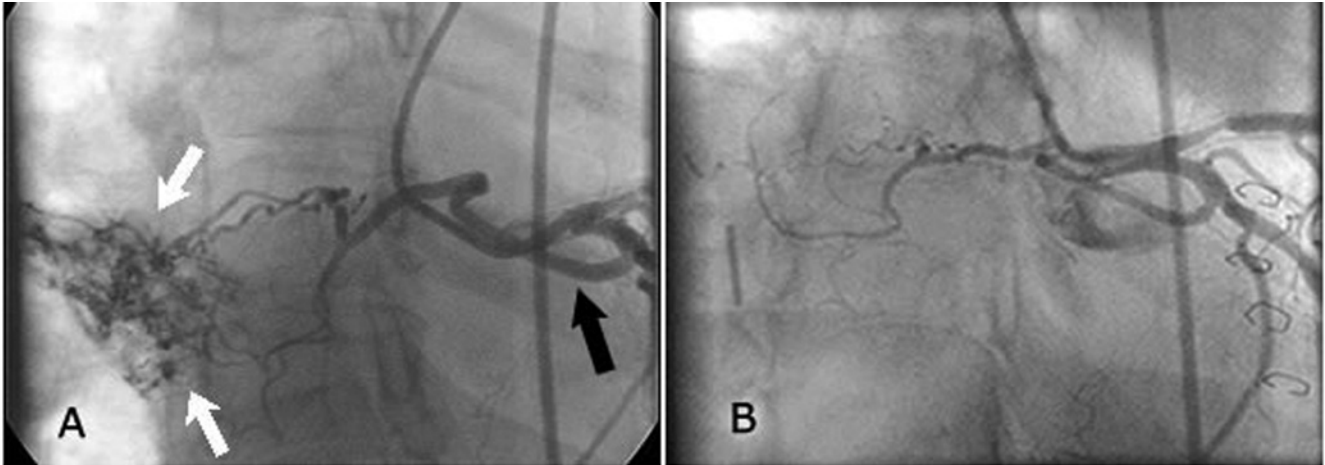


Figure 1. A, Preoperative angiography: the black arrow points to the aberrant artery arising from the proximal part of the circumflex artery; the white arrows point to the vascular network of the sequestered, hypoplastic mid-lobe. B, Postoperative angiography where the vascular network is no longer detected.

medically managed. Hung reported a case of bilateral pulmonary sequestration with blood supply originating from a coronary fistulous vessel arising from the circumflex artery and draining into the pulmonary artery [Hung 1996]. Hilton and colleagues reported a pulmonary sequestration supplied by several coronary arteries, including the circumflex artery [Hilton 1995]. No surgical therapy was given. Nakayama and associates described a case of right lung sequestration receiving arterial supply from the circumflex artery with myocardial ischemia caused by vasospastic angina and stealing from the coronary circulation

[Nakayama 2000]. The patient, a 48-year-old man, underwent surgical resection of the feeding artery without resection of the pulmonary sequestration. He received medical treatment for vasospastic angina.

In our patient, recurrent pulmonary infections and x-ray findings typical of the disease were absent. Congenital hypoplasia of the mid-lobe may explain in part the absence of recurrent pulmonary infections. Sequestration was coincidentally diagnosed during catheterization and confirmed by computed tomography and magnetic resonance angiography. The history of hemoptysis could be related to pulmonary pathology,

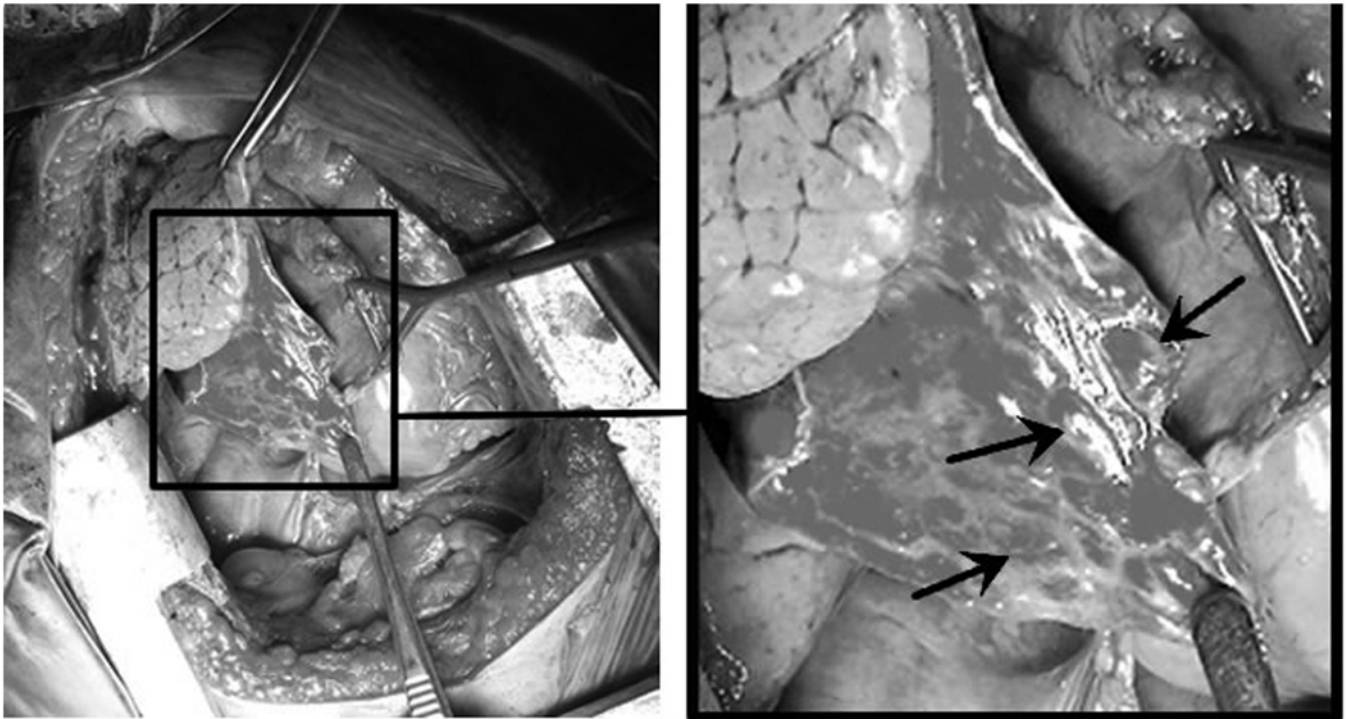


Figure 2. Intraoperative view of the sequestered hypoplastic lobe. Arrows point to the cystic elements of the sequestration.

but pulmonary sequestration was not diagnosed during past hospitalizations. We chose to proceed with combined myocardial revascularization and resection of the sequestered lobe to avoid recurrence of hemoptysis and eventual stealing from the coronary circulation. Although it would be technically rational to perform myocardial revascularization and proceed with lobe resection, we chose to go first with lobectomy to avoid the risk of bronchial bleeding that may occur during total heparinization required in cardiopulmonary bypass. Pre-bypass clamping of the bronchus could prevent this eventuality, but there was uncertainty about the exact course of the feeding vessels. Moreover, there was no evidence of pulmonary infection and eventual contamination was unlikely to occur. The location of the sequestration at the right middle lobe allowed unrestricted surgical access through median sternotomy. Although combined bypass surgery and resection of sequestered pulmonary segments supplied by the coronary arteries (mainly the right coronary artery) have been described previously [Morii 2002; Alvarez Suero 2004], to our knowledge this is the first case of sequestered mid-lobe hypoplasia supplied by the circumflex artery and associated with coronary artery disease that was successfully treated by a 1-stage surgical approach.

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