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Two unusual variants of pulmonary intra-lobar sequestration

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Abstract

A pulmonary sequestration is a congenital malformation characterized by non-functional lung tissue with abnormal arterial systemic supply and abnormal connection to the bronchial tree. This may lead to recurrent infections rendering a surgical intervention more demanding. Because of multiple anatomic variations, it is important to obtain high-quality preoperative radiological clarification to determine the most suitable surgical approach. Although a non-surgical technique, consisting of embolization of the aberrant artery has been described, a surgical technique remains the treatment of choice in operable patients. Preoperative embolization of the aberrant artery may reduce the risk of haemorrhage but could cause technical challenges in a hybrid approach and therefore unforeseen perioperative stress to the surgical team. We report 2 adult patients with unusual intra-lobar sequestration with aberrant vascular rare anatomy. Both were treated by surgery. In the latter patient, we performed a hybrid approach. This was complicated by perioperative coils exposure making it a technical challenge to proceed.

Keywords: Pulmonary sequestration • Embolization • Lobectomy

INTRODUCTION

Bronchopulmonary sequestration is a rare congenital disease characterized by non-functional dysplastic lung tissue with an anomalous systemic arterial supply and no macroscopical connection to the tracheobronchial tree [1].

It was first described by Pryce in 1946 and classified into 3 subtypes. Type I is a normally connected lung vascularized by an abnormal artery. Type II is characterized by a sequestered mass within the normal lung and an abnormal artery supply. Type III designates a sequestered mass outside of the normal lung vascularized by an abnormal artery as well [2].

Nowadays, 2 types of sequestration are described within the literature. An extra-lobar sequestration (25%) has its own pleura and venous drainage into the systemic veins (azygos vein or vena cava), meanwhile an intra-lobar sequestration (75%) is included in the visceral pleura of the normal lung with venous drainage into one of the pulmonary veins [1].

An intra-lobar sequestration usually gets his arterial supply from the thoracic (86.1%) or abdominal (6.9%) aorta. There is a left-sided preponderance [3].

Next to incidental computed tomography (CT) findings, the most common clinical presentation is recurrent infection.

Surgery is currently the preferred option of choice in symptomatic patients. Embolization of the systemic arterial blood supply has been described as a sole or neoadjuvant treatment. Preoperative embolization can occur by coils, vascular plug or

non-metallic substances like glue, polyvinylalcohol or polymers, and reduces the risk of haemorrhage [4]. However, this hybrid approach also can cause some perioperative difficulties as previously reported by Nakanishi *et al.* [5].

CASE PRESENTATION

We describe 2 patients with either a rare variant of arterial supply or an unusual venous drainage.

Our first patient was a 36-year-old male presenting recurrent respiratory infection. A computed tomography angiography showed an intra-lobar sequestration within the left lower lobe (LLL). The arterial supply was provided by a large trunk (17 mm × 20 mm) from the descending thoracic aorta dividing into 4 separate branches entering the sequestration (Fig. 1A). The venous return was also abnormal with drainage into the azygos vein (Fig. 1B). The patient underwent left lower lobectomy. Postoperative recovery was uneventful.

The second patient was a 40-year-old male presenting recurrent cough and bronchorrhea. A computed tomography angiography showed an infected intra-lobar sequestration of the right lower lobe (RLL) supplied by a systemic infra-diaphragmatic systemic branch arising from the left gastric artery (Fig. 2) with usual venous drainage into the left inferior pulmonary vein. Because of this unusual arterial supply with a risk of uncontrollable perioperative bleeding, we decided to proceed with a hybrid technique.



Figure 1: (A) Volume rendering technique angio-CT of the large arterial supply of LLL sequestration from descending aorta. (B) Axial thoracic CT in mediastinal window of the azygos drainage of LLL sequestration.

Preoperatively the patient underwent embolization of the aberrant artery by metallic coils by the interventional radiologist after which we took the patient into the operating theatre to perform video-assisted lobectomy of the right lower lobe. During the surgery, we encountered some difficulties in transecting the aberrant artery due to distal migration of the metallic coils leading up to intrathoracic visualization of these coils after the use of the endoscopic stapler device. There was minor bleeding controlled by adding clips. Postoperative recovery was uneventful.

CONCLUSION

There are many vascular variants of pulmonary sequestrations. Most patients undergo surgical treatment. In this report, we describe an anomalous venous drainage into the azygos vein in an intra-lobe sequestration, where we expect normally drainage into the pulmonary vein. Imaging showed an unusual large feeding branch from the aorta. Our second case showed an infra-diaphragmatic systemic arterial blood supply from the left gastric artery, treated preoperatively by embolization. Due to migration of the coils a transection of this aberrant artery was difficult.

Preoperative vascular imaging is of utmost importance to determine the exact anatomy for surgical planning and prevent potential intraoperative haemorrhage. Embolization preoperatively may be used to reduce the risk of perioperative bleeding, but one should be aware of possible migration of the coils rendering a resection more difficult. Its indication should be preserved for highly selected cases. We would like to emphasize the potential danger of using metallic coils preoperatively. The use of non-metallic agents distally in addition to a proximal vascular plug might be a valid alternative.

Conflict of interest: none declared.

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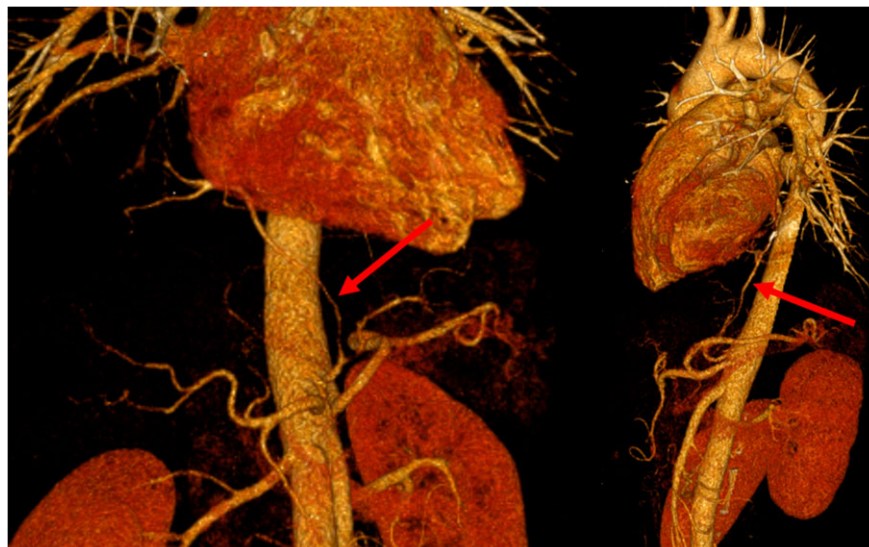


Figure 2: Volume rendering technique angio-CT of the systemic arterial supply of RLL sequestration from coeliac trunk variant passing through oesophageal hiatus.

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