Emergency laparotomy helped the resection of an intralobar pulmonary sequestration with haemorrhagic shock

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Abstract

Massive intrapulmonary haemorrhage and haemothorax are uncommon presentations associated with pulmonary sequestration. Here, we describe the case of a 40-year-old man who suffered from high fever and haemoptysis for 1 week before he was admitted to our hospital with a complaint of chest discomfort with shock. Computed tomography revealed that pulmonary sequestration supplied from the coeliac artery with persistent bleeding. The patient underwent right lower lobectomy and an emergent laparotomy for ligation of the aberrant artery. A pulmonary sequestration has a severe complication resulting in shock due to intrapulmonary haemorrhage and haemothorax. Accordingly, early resection of a sequestered lung should be the choice of the treatment in these cases.

Keywords: Intralobar sequestration• Haemothorax• Haemorrhagic shock• Emergency

INTRODUCTION

Sequestrations are rare pulmonary malformations that do not communicate with the normal bronchial system and have their own blood supply mostly from the aorta [1, 2]. Pulmonary sequestrations are divided into two groups: extralobar sequestration and intralobar sequestration (ILS). Approximately 75% of all cases in the literature are ILS. ILS is included within the visceral pleura located mostly in left lower lobe [3]. Recurrent pulmonary infection and haemoptysis are representative symptoms. However, massive intrapulmonary haemorrhage and haemothorax are rarely reported as presenting symptoms.

CASE

Owing to right chest pain and dyspnoea, a 40-year-old man without any past illness was admitted to our emergency department. His physical parameters at admission were as follows: body temperature, 39.1°C; pulse rate, 138 per min; blood pressure, 110/70 mmHg and oxygen saturation, 93% in air. His initial laboratory test yielded a white blood cell count of 22 900/µl, haemoglobin level of 8.1 g/dl and C reactive protein level of 34.9 mg/dl. Chest radiography showed a large mass in the right lower field with the mediastinum shifted to the left. With a chest tube insertion at the emergent department, an enhanced chest computed tomography (CT) was performed. Massive haemorrhage within the right lower lobe and right haemothorax were observed; in addition, an aberrant artery of diameter 10 mm originating from the coeliac artery was identified (Fig. 1). The patient presented with shock due to active bleeding caused by a ruptured anomalous artery.

General anaesthesia was performed at the seated position because of persistent respiratory discomfort. Preoperative haemoglobin level dropped to 6.1 g/dl. Upon open thoracotomy, we attempted right lower lobectomy in the left lateral decubitus position. However, the right lower lobe was so distended that we could not continue the procedure (Fig. 2). Therefore, we performed laparotomy after a postural change to the supine position and ligated the aberrant vessel below the diaphragm by using an autosuturing device (Endo GIA™, Covidien Japan). Right lower lobectomy was then carried out. The right upper and middle lobes were small and appeared hypoplastic. The post-operative course was uneventful, and the patient was discharged home on the 14th day after the operation.

DISCUSSION

Unlike other cases of sequestration, our case was extremely emergent and the patient presented with massive intrapulmonary haemorrhage with shock. Savic et al. reported that the first symptoms of ILS occur before the age of 10 in 37.2% of all patients. In 15.5% of cases, the disease remains asymptomatic and is diagnosed by chance. The most common symptoms are cough, expectoration and recurrent respiratory infection, all of which become more severe with time [3]. Haemothorax and intrapulmonary haemorrhage caused by pulmonary sequestration are rather rare presentations [1, 4]. Pryce reported for the first time in 1946 the classification of sequestration according to the distribution of an aberrant
artery [5]. A sequestration has its own feeding artery originating from a systemic artery. In most cases, ILS is supplied by the descending aorta. However, the abdominal aorta feeds the sequestered lung less frequently than the descending aorta. Although less common, the intercostal artery, subclavian artery, internal thoracic artery, innominate artery, coeliac artery and splenic artery can also be the origin of a feeding artery. In their series, Savic et al. [3] showed that 15% (55/373) of all cases had more than one aberrant artery. Angiography is still a useful procedure for diagnosing and evaluating sequestrations. Recently, spiral CT angiography has been used as a particularly effective and less invasive technique and has the potential to be the first choice in the diagnosis and assessment of pulmonary sequestration; however, several reports state that small anomalous vessels <5 mm in diameter can be difficult to detect using this method [4].

Resection of the malformation is advocated in all cases where complications occur. However, whether an asymptomatic case is an indication for operation is still controversial. Understanding of aberrant vessels is mandatory prior to resection of sequestration. Although aberrant arteries are sometimes easy to identify within the pulmonary ligament, there are usually inflammatory changes in the area of the sequestration because of recurrent infections; moreover, the arteries are hidden in the scarred tissue. Regarding the ligation of aberrant vessels, stapling devices can be used safely even for vessels with large diameters. In our operative experience, although adhesion was so hard, the preoperative understanding of the localization of the aberrant artery was rather helpful for the lobectomy, and ligation using the stapling device was both safe and successful.

In addition to surgical ligation, interventional embolization is considered a feasible method of controlling bleeding from aberrant vessels. As far as the current case is concerned, we believe that interventional angiography is not a safe method in case of shock. Given a successful embolization, additional laparotomy may be avoidable. However, considering incomplete embolization and the patient’s condition such as persistent respiratory discomfort, surgical treatment was thought to be inevitable.

Considering that the patient had a cold and haemoptysis since a week before admission, the cause of the bleeding was thought to be the collapse of the aberrant vessels. However, it was too difficult to detect the bleeding point pathologically because the sequestered lung was filled with large amount of blood.

To our knowledge, this is the first case report that shows that open laparotomy is rather helpful in the surgical treatment of pulmonary sequestrations. We think it reasonable that additional laparotomy was inevitable because the sequestrated lung was so extended that it was impossible to mobilize and use any thoracoscopic instruments. Intrapulmonary haemorrhage and haemothorax are a rare but life-threatening complication of a pulmonary sequestration. The resection of the pulmonary sequestration is recommended in all cases to prevent further complications.

Conflict of interest: none declared.
REFERENCES


