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Case report

Endovascular embolization prior to surgical resection of symptomatic intralobar pulmonary sequestration in an adult

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ABSTRACT

Intralobar pulmonary sequestration is a rare congenital malformation, conventionally managed by surgical resection. Recently, the endovascular embolization has been proposed for the definite treatment of this disease. Additionally, preoperative embolization of aberrant arteries to minimize the risk of serious intraoperative haemorrhage has also been described. We report the case of 43-year old female patient who presented with cough and haemoptysis, and was successfully treated with endovascular embolization followed by a Video-assisted thoracoscopic wedge resection.

1. Introduction

Intralobar pulmonary sequestration (ILS) is a rare malformation characterised by non-functioning lung tissue, separated from the tracheobronchial tree. ILSs receive a systemic artery supply from the descending thoracic aorta, abdominal aorta, the celiac trunk or from intercostal arteries [1]. Diagnosing ILS is based on imaging and identifying the systemic arterial supply. Haemoptysis and/or recurrent infections are present in two-thirds of patients [2,3]. The classical therapeutic approach is surgical resection in which the abnormal lung tissue is removed thus giving no opportunity for complications like ischemic infarction or abscess formation [4,5]. However, inadvertent injury of the aberrant artery can cause a severe and potentially life threatening haemorrhage during pulmonary resection. To overcome this problem, preoperative embolization of aberrant systemic arteries with metallic coils or vascular plugs, followed by surgical resection, has recently been proposed [6]. Additionally, the endovascular embolization as a definite therapy of ILS has also been described [7–9]. We present a case of an adult with ILS in whom endovascular embolization using a combination of metallic coils and vascular plugs was performed to allow a safe surgical resection.

2. Case report

43-year old female with history of asthma and recurrent respiratory infections presented with cough and massive haemoptysis. Physical findings, laboratory results and chest radiography were normal. To exclude pulmonary embolism, multidetector computed tomography angiography (CTA) was performed, revealing an area of consolidation with cystic components in posterior basal segment of the left lung. Two nutrient arteries were seen arising from the left side of the descending thoracic aorta, following a transverse course toward the abnormal lung parenchyma, with venous drainage via the inferior pulmonary vein (Fig. 1a). Despite the antibiotic and antitussic therapy, the patient eventually got worse and was transferred to thoracic surgery department. In collaboration with interventional radiologist, the endovascular treatment of ILS was proposed to control the bleeding. After local anaesthesia, a short 5 F introducer sheath (Terumo Europe N.V., Belgium) was put in place via the right transfemoral route. Prior to embolization, aortography confirmed the presence of the two nutrient arteries, which were selectively catheterized with a 5 F catheter (Sidewinder®, Terumo Europe N.V., Belgium) (Fig. 1c and d). The 0.035 stiff straight 260 cm long guidewire was left in the nutrient arteries and the short 5 F introducer sheath was replaced with a long (55 cm) 7 F introducer sheath (Cordis Corp., Miami, FL, USA). A 2.4 F microcatheter (Progreat®, Terumo Europe N.V., Belgium) was then superselectively positioned throughout 5 F Sidewinder catheter in the feeding arteries before embolization with coils. The small distal branch of the upper vessel was embolized with 2×3 mm pushable coils (VortX-18 fibered platinum coil, Boston Scientific, Cork, Ireland) and in the proximal part of the artery,

Abbreviations: ILS, intralobar pulmonary sequestration; CTA, computed tomography angiography; MRI, magnetic resonance imaging; VATS, video-assisted thoracoscopic surgery; AVP, Amplatzer Vascular Plug.

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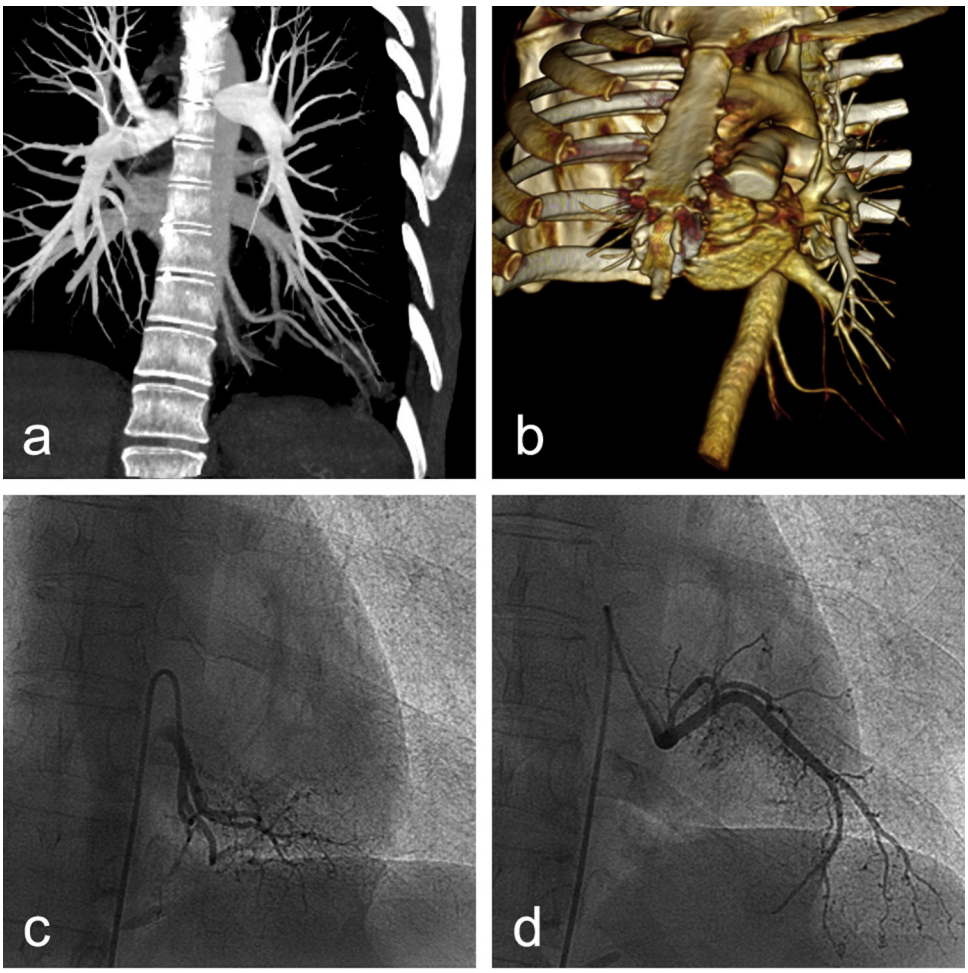

Fig. 1. Two nutrient arteries arising from descending thoracic aorta and transversing laterally toward consolidated lung parenchyma are shown on coronal maximum intensity projection (MIP) reconstruction (a) and on 3D volume rendered image (b); findings are consistent with intralobular pulmonary sequestration. Selective angiography confirmed the diagnosis, showing in detail the lower (c) and upper (d) aberrant artery.

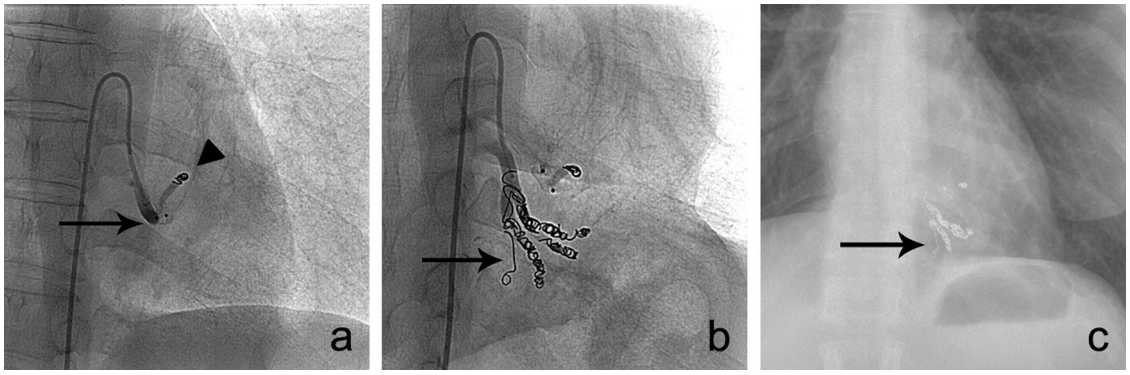

Fig. 2. Postembolization control confirms total occlusion of upper aberrant artery (a) with Amplatzer Vascular Plug (arrow) and coil (arrowhead). Lower artery (b) was successfully embolized with coils (arrows). Control chest radiography 10 days after procedure is normal (c); embolization material is seen in medial aspect of left basal lung (arrow).

the 6 mm Amplatzer Vascular Plug (AVP; AGA Medical, Plymouth, MN, USA) was deployed. We selected AVP approximately 30% larger than the vessel diameter for secure fitting, prevention of device migration, and total occlusion. Selective angiography of the lower artery revealed four distal branches, which were subsequently closed through microcatheter with 2×3 mm and 4 mm pushable coils (Vortex-18 fibered platinum coils, Boston Scientific, Cork, Ireland Boston). In the proximal part of the artery, the 6 mm detachable coil was used (Interlock-18, fibered platinum coil, Boston Scientific, Cork, Ireland Boston). The control angiogram confirmed successful occlusion of the aberrant arteries (Fig. 2). No incident or complication occurred during the procedure. During the hospitalization patient was observed in semiintensive care unit and no clinical symptoms were present. Antibiotics were administered during hospitalization to prevent any septic events and non-steroidal anti-inflammatory drugs were used for analgesia. The

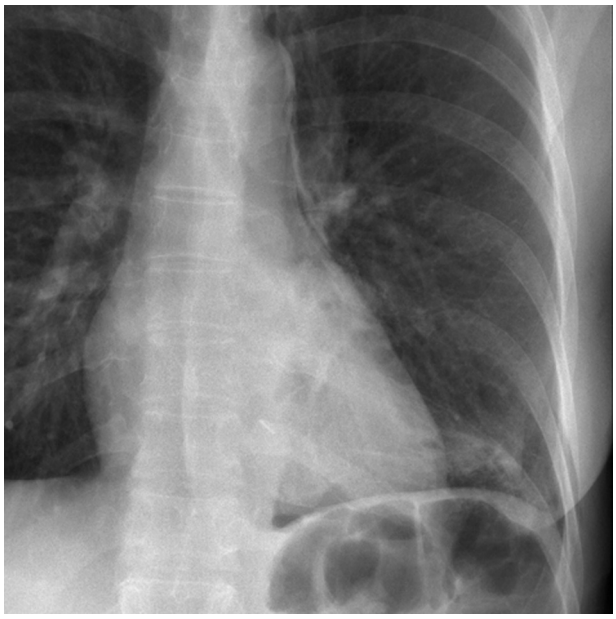

Fig. 3. Control chest radiography 3 months after surgery is normal.

patient was discharged from hospital after 3 days. In 4 weeks after the procedure, the patient was completely free of haemoptysis, dyspnea or respiratory infection, but reported occasional chest pain and recurrent cough. After preparation, elective Video-assisted thoracoscopic (VATS) pulmonary wedge resection was successfully performed and the patient uneventfully discharged home on postoperative day 2. Control chest radiograph 3 months after surgery showed no consolidation (Fig. 3) and at follow-up after 14 months, the patient was completely free of symptoms, reporting a clear improvement of her quality of life.

3. Discussion

Pulmonary sequestration is a relatively rare anomaly comprising 0.15–6.4% of all congenital pulmonary malformations [3,4]. A differentiation is made between intralobar sequestrations, which share a common pleura with the normal lung tissue, and extralobar sequestrations, which are separated from the remaining lung tissue by a separate lining of pleura [4]. ILSs are generally isolated congenital bronchopulmonary anomalies, preferentially affecting the posterior basal segment [4]. Sequestration occurs as a result of failure in obliteration of systemic vessels originating from early embryonic splanchnic vessels [7]. Masses of non-functioning lung parenchyma are contiguous with the normal adjacent lung tissue and separated from the normal tracheobronchial tree. On the left side the ILSs receive a systemic artery supply from the descending thoracic aorta, whereas on the right side it usually arises from the abdominal aorta or the celiac artery. Venous drainage is most commonly via pulmonary veins. The majority of patients are asymptomatic, however recurrent pulmonary infection, cardiac failure due to left-to-left shunt and haemoptysis are possible clinical presentations [2,3].

Thoracic CTA is the most useful test in the evaluation of patients with suspected abnormal systemic arterial supply to the lung, as it demonstrates both the bronchial and vascular anatomy of the lung. Computed tomography may reveal a focal area of ground glass density, indicating an area of relative hypervascularity and/or intra-alveolar haemorrhage [2,3]. However, magnetic resonance imaging (MRI) or angiography can also be used to demonstrate aberrant vasculature and bronchial anatomy. Chest radiography is not pathognomonic and may reveal a pneumonia-like pulmonary consolidation or a soft tissue mass with well defined edges and air-fluid levels [1–3].

The gold standard for treatment of ILS is a surgical resection, which is indicated for all patients because of a potential risk for recurrent severe infections, massive haemoptysis, and congestive heart failure caused by left sided cardiac overload [4,5]. However, resection by thoracotomy may result in additional morbidity as well as aesthetic sequelae. This is especially true in paediatric population, because it causes skeletal and muscle deformities, such as asymmetry of the chest wall, scoliosis or winged scapula. When possible, these complications may be reduced by minimally invasive VATS. The surgical approach relies on identifying and controlling aberrant arterial supply. If all the arteries are not properly controlled, life threatening haemorrhage may occur [5,6].

Endovascular embolization of intralobar pulmonary sequestration has been reported as a safe alternative to surgery since 1998, but gained wider acceptance mostly as an alternative treatment option in paediatric patients [8]. Data for the endovascular embolization in adult symptomatic patients are sparse [7,9,10]. Major concerns include possible incomplete occlusion of arterial supply, resulting from distal embolization of feeding vessel with subsequent opening of collateral supply. This may lead to evolution of the sequestered tissue and possible recurrence of symptoms and infection. Distant migration of embolization material, resulting in embolization of non-targeted arteries has also been reported [7]. Metallic coils and AVPs represent the preferred embolization material in a majority of previously reported cases. Studies, mostly case series confirm that endovascular embolization is a successful treatment strategy [7,9,10]. In addition, preoperative embolization has recently been proposed to prevent intraoperative bleeding [8]. The aberrant artery needs to be identified and controlled early during the surgical resection and potential injury to a systemic artery, which may be friable due to chronic inflammation, can lead to massive haemorrhage [8]. Preoperative embolization obliterates the major arterial supply to sequestrated lung tissue, thus making the VATS procedure much easier and safer.

Our patient presented with cough and haemoptysis and was successfully treated with endovascular embolization and surgical resection. Due to the small caliber of arteries we decided to use the combination of pushable and detachable coils and AVPs to minimize the risks of regurgitation and non-target occlusion and to enable faster embolization. No complication related to vascular access or embolization occurred during the endovascular procedure and the following surgical resection 4 weeks later was successful. Our case shows that embolization as a definite treatment or as a preoperative procedure can be performed in a safe and effective manner. We hope that our encouraging results will stimulate other tertiary centers to organize similar multidisciplinary approach for evaluation of patients with ILS with an algorithm that includes endovascular embolization.

4. Conclusion

The present case report describes the successful clinical outcome of endovascular and surgical treatment of a patient with symptomatic ILS. We conclude that endovascular embolization is a safe and feasible treatment option, with alternative role as a preoperative procedure for ILS in adults.

Conflict of interest

All authors declare that they have no conflicts of interest.

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