Use of Embolization and Surgical Resection in a Case of Pulmonary Sequestration Manifesting with Hemoptysis

Hemoptizi ile Seyreden Pulmoner Sekestrasyonlu bir Olguda Embolizasyon ve Cerrahi Rezeksiyon Uygulaması

Orhan Yücel, Hasan Çaylak, Alper Gözübüyük, Kuthan Kavaklı, Onur Genç

Department of Thoracic Surgery, Gülhane Military Medical Academy, Ankara, Turkey

ABSTRACT

Pulmonary sequestration is the term used to describe a rare embryonic non-functional lung tissue which has no bronchial communication with the normal tracheobronchial tree. In this study, we reviewed a patient who was treated with embolization as a conservative treatment with a coil first, and after three months underwent excision of extralobar sequestration via right thoracotomy because of his continuing complaints of hemoptysis. We conclude that the embolization can be a conservative treatment option, and the definitive treatment of pulmonary sequestration is surgical resection to avoid destruction of the normal pulmonary parenchyma due to infection. *(Tur Toraks Der 2011; 12: 172-5)*

Key words: Bronchopulmonary sequestration, hemoptysis, embolization, thoracic surgery

Received: 03.05.2009 *Accepted: 21.05.2009*

ÖZET

Pulmoner sekestrasyon terimi normal trakeobronşiyal sistem ile bronşiyal bağlantısı olmayan, nadir görülen ve fonksiyonel olmayan akciğer dokusu için kullanılır. Bu çalışmada, ilk olarak coil embolizasyon ile konservatif tedavi uygulanan ve hemoptizi şikayetinin devam etmesi üzerine üç ay sonra ekstralobar sekestrasyon rezeksiyonu yaptığımız bir olguyu gözden geçirdik. Sonuç olarak embolizasyonun sadece konservatif tedavi seçeneği olabileceğini ve normal akciğer parankim dokusunun enfeksiyona bağlı harabiyetini önlemek için kesin tedavinin rezeksiyon olduğunu düşünmekteyiz. *(Tur Toraks Der 2011; 12: 172-5)*

Anahtar sözcükler: Bronkopulmoner sekestrasyon, hemoptizi, embolizasyon, torasik cerrahi

Geliş Tarihi: 03.05.2009 *Kabul Tarihi: 21.05.2009*

INTRODUCTION

Pulmonary sequestration (PS), a rarely seen congenital malformation, is primitive lung tissue without any connection with the tracheobronchial tree. Abdominal aorta, intercostal artery, internal mammarian artery, subclavian artery and circumflex coronary artery can be the source of systemic blood circulation but the most common is the thoracic aorta [1]. While variable drainage may be to the superior and inferior cava, brachiocephalic or azygous veins, the common venous drainage is to the pulmonary vessels [1]. The sequestrations can be in two forms: the first is intralobar sequestration (ILS) where the sequestered part of the lung lies within normal pulmonary visceral pleura; and the second is extralobar sequestration (ELS) where the abnormal segment of lung is completely separate and enclosed in its own pleural investment. A solid or cystic mass in the base of one lung is the usual radiographic finding [1,2]. In this article, we reported a patient having pulmonary sequestration manifesting with hemoptysis, reviewed the literature and discussed the treatment of embolization and resection.

CASE

A 32-year old man was admitted to hospital with the complaint of hemoptysis which was not massive. Respiratory tract infection was detected in our case and medical treatment was administered. Computed tomography (CT) showed a pulmonary cystic lesion containing air and fluid (Figure 1). Firstly therapeutic embolization as a conservative treatment was achieved with a coil because the patient did not receive surgical treatment (Figure 2). Then, the patient was discharged from hospital after embolization After three months, he was admitted again complaining of hemoptysis, but he had also had progressive back pain after embolization. We planned to operate and he was successfully treated with an excision of ELS via left thoracotomy (Figure 3). The blood supplies to the sequestration were from the seventh thoracic vertebra level of the thoracic aorta with four vessels while venous drainage was via the inferior pulmonary vein. No postoperative complications were observed. The patient was asymptomatic in the 14-month follow-up.

Address for Correspondence / Yazışma Adresi: Kuthan Kavaklı, Department of Thoracic Surgery, Gülhane Military Medical Academy, Ankara, Turkey
Phone: +90 312 304 51 71 E-mail: dr_kuthan_78@hotmail.com

doi:10.5152/ttd.2011.40

DISCUSSION

Allthough ILS generally produces symptoms, mostly respiratory tract infection, ELS may rarely be asymptomatic. ELS is most commonly shown in the left hemithorax and is often discovered incidentally. Chest roentgenogram is generally not pathognomonic. It is revealed that a pulmonary consolidated lesion simulates pneumonia, or a soft tissue mass with well or ill defined edges and air-fluid levels, which can also be seen [1]. The definitive diagnostic tool of ELS is selective angiography. However, the feeding vessels can be determined by contrast-enhanced CT, as in our case (Figure 1). Selective angiography exposes the aberrant arterial supply and venous drainage of the sequestration. At this point, excluding differential diagnoses such as indeterminate cystic lesion, foreign bodies or tumors will be very helpful. In order to guide an effective surgical treatment procedure, planning aortic and pulmonary arterial angiographic studies is very important.

Figure 1. Parenchymal image of CT which shows the pulmonary cystic lesion and visceral pleura of the sequestrate lobe

It was reported that 25% of sequestrations are ELS which are triangular and occur in the left chest 90% of the time [2]. It is usually found in the posterior costophrenic angle, but rarely occurs in the mediastinum or within or beneath the diaphragm in a periadrenal location [1,2]. In our case, ELS was localized in the left hemithorax. Usually ELS is not solitary, it is commonly associated with congenital malformations including congenital heart disease, congenital cystic adenomatoid malformation, chest wall and vertebra-diaphragm abnormalities. However, in our case, there weren't any additional congenital malformations [1].

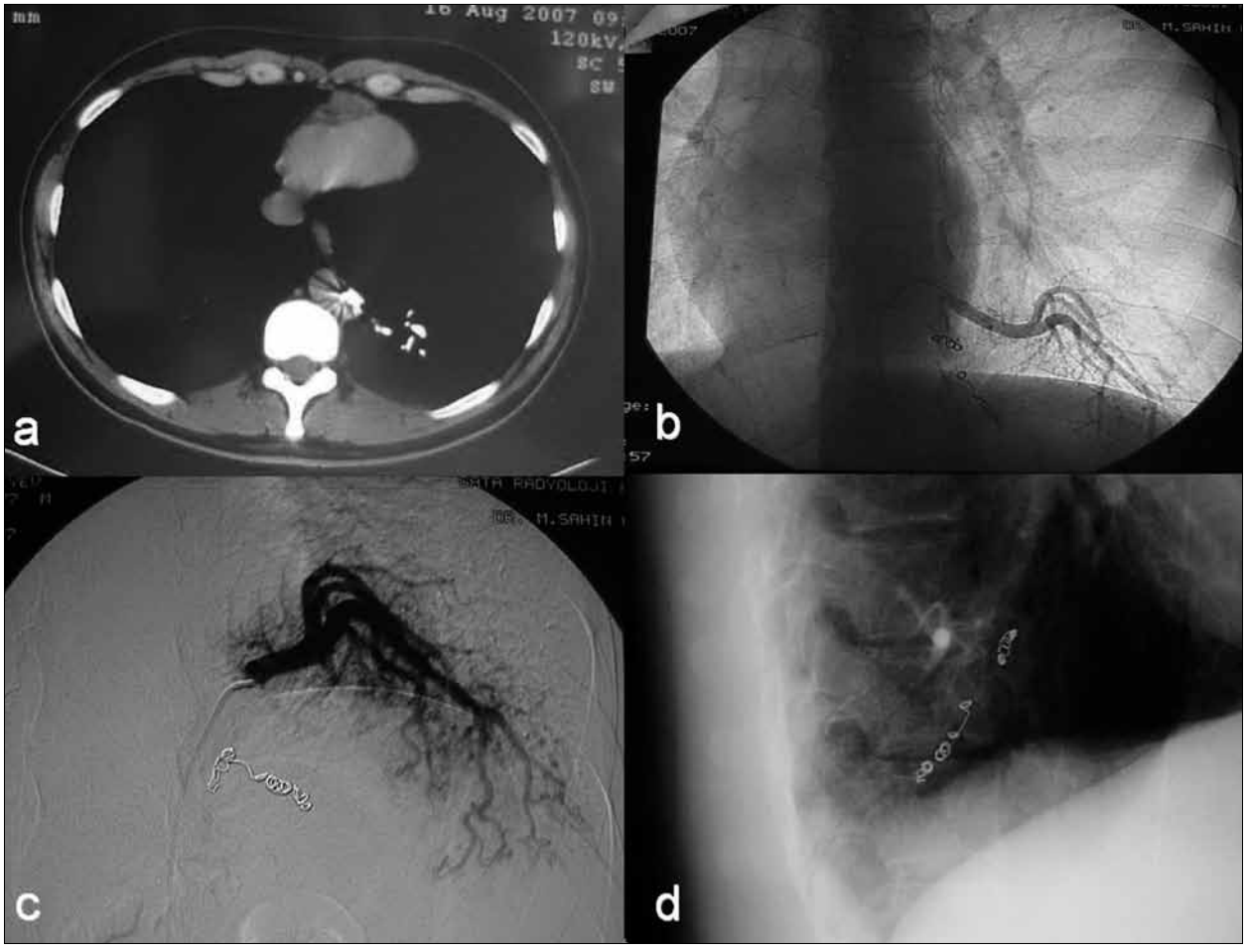

Figure 2. Axial plain of CT of the thorax reveal a feeding vessel of the sequestration arising from the thoracic aorta (a). Aortagraphy shows the feeding vessels and coil therapeutic embolization is performed (b, c). Lateral chest roentgenogram shows the coils which were used for therapeutic embolization 4-month earlier to treat the hemoptysis

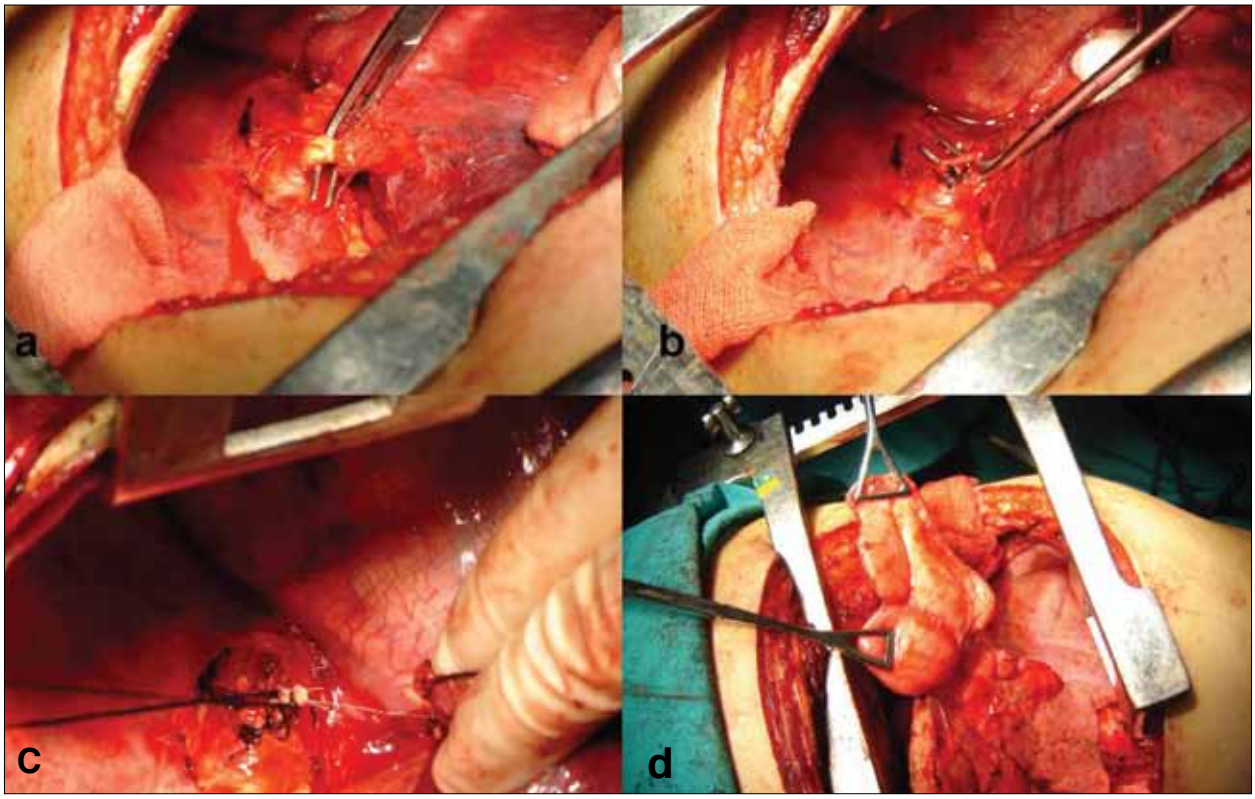

Figure 3. Major feeding vessel of the sequestration which was discovered after careful dissection of the inferior pulmonary ligament (a). Fourth feeding vessel of the sequestration is shown after being tied with silk (b). The coil which was used for therapeutic embolization was found when the last feeding vessel was divided (c). ELS visseral pleura is shown (d)

Since the artery of sequestration arises from a systemic artery, to supply the pulmonary sequestration, this artery often arises from the abdominal aorta that penetrates the diaphragm [3]. An abnormal artery, which originates from the descending aorta is almost always single [1,3]. Whereas the diameter of the artery is generally between 0.5 and 2 cm, if the arterial diameter is observed as smaller than 3 mm, it is thought more likely to be a component of multiple arteries [1]. Venous drainage is almost always via the inferior pulmonary vein. In our case, the arterial supply was from the thoracic aorta while the venous drainage was through the pulmonary vein. In our previous studies, we observed that sequestration is generally seen in the lower lobes and the abnormal artery inside the pulmonary ligament [1]. In addition, the fragile elastic structure of the abnormal artery makes it more susceptible to damage [4]. For this reason, during surgery, the inferior pulmonary ligament should be carefully dissected to avoid injury to the aberrant artery existing within it. On the other hand, morbidity and even mortality may occur as a result of massive hemorrhage because of infection due to the air transfer between the normal pulmonary parenchyma and sequestration area, which allows bacterial infections following bacterial transfer by way of microscopic connections. Extralobar sequestration with hemoptysis is a very rare presentation and, according to our knowledge, there is only one case presentation in the literature [5]. Herein, we presented the second case of extralobar sequestration presenting with hemoptysis.

The treatment of choice has always been surgical excision. Most authors consider that the bronchopulmonary sequestration should be treated with ligation of the afferent artery and resection of the sequestered pulmonary parenchyma due to the propensity for recurrent infections [1,2,6]. Coil embolization is a new and a less invasive technique than surgical treatment. Transarterial embolization is an accepted treatment for hemoptysis The indications for coil embolization are various. The method can be used to eliminate right-to-left shunting of blood in arteriovenous fistulas, protect the lungs from excessive intraoperative bronchial blood flow, eliminate left ventricular volume overload, prevent collateral blood flow, and to occlude unwanted shunts [7]. Only a few reports have described embolization as the sole treatment for this disorder. Hayakawa et al. have performed transarterial embolization in patients with massive hemoptysis due to sequestration [8]. However no long-term results are available in the literature. As pointed out, this pulmonary embolization can be regarded as a conservative treatment method in order to control of symptom of hemoptysis. In our case, we initially suggested the surgical treatment but the patient did not accept surgery and we performed embolization. Because of his continuing symptom of hemoptysis, we performed resection as a definitive treatment.

Even though the use of therapeutic embolization is obviously very essential, we had a case for whom it was not sufficient to control hemoptysis. As we pointed out in our previous studies, optimal treatment of pulmonary sequestration is surgical resection, especially in symptomatic cases with hemoptysis [1,3,9,10].

REFERENCES

1. Yucel O, Gurkok S, Gozubuyuk A, et al. Diagnosis and surgical treatment of pulmonary Sequestration. Thoracic Cardiovascular Surg 2008;56:154-7. [CrossRef]
2. Gezer S, Tastepe I, Sirmali M, et al. Pulmonary sequestration: a single-institutional series composed of 27 cases. J Thorac Cardiovasc Surg 2007;133:955-9. [CrossRef]
3. Savic B, Birtel F J, Tholen W, et al. Lung sequestration: report of seven cases and review of 540 published cases. Thorax 1979;34:96. [CrossRef]
4. Genç O, Gürkök S, Dakak M, et al. Pulmonary sequestration and surgical treatment. Asian Cardiovasc Thorac Ann 2006;14:3-6.
5. Sato Y, Endo S, Saito N, et al. A rare case of extralabar sequestration with hemoptysis. J Thorac Cardiovasc Surg 2004;128:778-9. [CrossRef]
6. Halkic N, Cuenoud PF, Corthesy ME, et al. Pulmonary sequestration: A review of 26 cases. Eur J Cardiothorac Surg 1998;14:127-33. [CrossRef]
7. Fuhrman BP, Bass JL, Castaneda-Zuniga W, Amplatz K, Lock JE. Coil embolization of congenital thoracic vascular anomalies in infants and children. Circulation 1984;70:285-9. [CrossRef]
8. Hayakawa K, Soga T, Hamamoto K, et al. Massive hemoptysis from a pulmonary sequestration controlled by embolization of aberrant pulmonary arteries: Case report. Cardiovasc Intervent Radiol 1991;14:345. [CrossRef]
9. Gurkok S, Gozubuyuk A, Dakak M, et al. Extralobar pulmonary sequestration with anomalous blood supply from the subclavian artery. The Internet Journal of Thoracic and Cardiovascular Surgery 2004;6.
10. Kaya S, Yıldırım Z, Biber C, et al. Intrapulmonary sequestration diagnosed preoperatively by means of angiography. Turkiye Klinikleri J Med Sci 1996;16:172-4.