



# Thoracoscopic treatment of pulmonary sequestration<sup>☆</sup>

Peter B. Kestenholz, Didier Schneiter, Sven Hillinger, Didier Lardinois, Walter Weder<sup>\*</sup>

*Department of Surgery, Division for Thoracic Surgery, University Hospital Zurich, Rämistrasse 100, CH 8091 Zurich, Switzerland*

Received 24 September 2005; received in revised form 31 January 2006; accepted 3 February 2006

## Abstract

**Objective:** Pulmonary sequestration is a rare congenital malformation and may be the cause of recurrent infections or hemoptysis. It has been shown in case reports that resection by video-assisted thoracic surgery (VATS) is feasible despite the possible technical difficulties due to inflammatory changes, but its role has not been evaluated yet in a larger series of consecutively treated patients. **Methods:** Retrospective analysis of all consecutively thoracoscopically treated patients (between January 1991 and January 2005) with pulmonary sequestration in a single center. We included 14 patients in the study who fulfilled the criteria; seven were women. Median age was 33 years (20–64 years). The following data were analyzed for all patients: major symptoms, diagnostic procedures, treatment, and outcome. Operative parameters and findings including operating time, blood loss, anatomical location of the sequestration, and feeding vessels were evaluated. **Results:** Leading symptoms were recurrent infections (10), hemoptysis (3), and chest discomfort (1). The diagnosis was made by CT scan. Additionally, an arteriography or an angio-MRI was done in three patients and one patient, respectively. Thirteen intralobar (all lower lobes, eight on the right) and one left-sided extralobar pulmonary sequestration were resected. We performed eight lobectomies, four atypical segmentectomies, one extralobar resection, and one occlusion of the aberrant artery. One case had to be converted to a thoracotomy due to bleeding from the aberrant artery. There was no mortality. Complications included pneumonia in three cases, one hemothorax, one pneumothorax after removing the chest tube, and one wound infection. All were treated conservatively. **Conclusion:** Thoracoscopic treatment of pulmonary sequestration is feasible in experienced hands. The aberrant systemic artery can be freed and dissected safely despite the frequently occurring inflammatory changes. Conversion rate to thoracotomy is low.

© 2006 Elsevier B.V. All rights reserved.

**Keywords:** Pulmonary sequestration; Treatment; VATS; Thoracoscopically

## 1. Introduction

Pulmonary sequestration is a rare congenital malformation. It represents 0.15–6.4% of all pulmonary malformations [1,2]. It is characterized by a mass of non-functioning pulmonary tissue and a lack of a normal communication with the tracheobronchial tree. Two types of pulmonary sequestration are recognized: intralobar sequestration, which is an abnormal region within the normal pulmonary parenchyma without own pleural covering, and extralobar sequestration with an own pleural covering. Pulmonary sequestration is vascularized by an aberrant systemic artery, most frequently from the descending thoracic or abdominal aorta. In intralobar disease the venous drainage is usually the pulmonary vein, and in extralobar disease it drains via the pulmonary vein or a systemic vein.

In symptomatic patients with pulmonary sequestration it is generally accepted to do resection.

The aim of this study was to evaluate the role of the approach by video-assisted thoracic surgery (VATS) in respect to feasibility, safety, and complications.

## 2. Patients and methods

We retrospectively reviewed the files of all patients diagnosed with pulmonary sequestration and treated by a thoracoscopic procedure at the division of thoracic surgery in the University Hospital of Zurich. In the period between January 1991 and January 2005, 14 adult patients were operated for a pulmonary sequestration. This included seven women and seven men. The median age was 33 years (range 20–64 years). The following data were collected from the hospital chart and during follow-up controls for all patients: major complaint resulting in the diagnosis, diagnostic procedures, treatment, and outcome. Operative treatment and findings including operating time, blood loss, anatomical location of the sequestration, and feeding vessels were evaluated.

<sup>☆</sup> Presented at the joint 19th Annual Meeting of the European Association for Cardio-thoracic Surgery and the 13th Annual Meeting of the European Society of Thoracic Surgeons, Barcelona, Spain, September 25–28, 2005.

<sup>\*</sup> Corresponding author. Tel.: +41 44 255 88 02; fax: +41 44 255 88 05.  
 E-mail address: [walter.weder@usz.ch](mailto:walter.weder@usz.ch) (W. Weder).

### 2.1. Surgery

The VATS procedures were performed under single lung ventilation in a full lateral position. We used three 10 mm trocars and one additional 5 mm trocar as described earlier in detail [3]. First, the aberrant artery was dissected, closed and cut with a stapling device (Multifire Endo GIA 30, 2.5 mm; Auto Suture, Tyco Healthcare, Norwalk, CT, USA). Afterwards a standard thoracoscopic lobectomy [3], wedge resection, or a resection of the extrapulmonary sequestration was performed. At the end of the procedure one trocar incision in the submammary fold was enlarged to 5–8 cm to remove the specimen.

## 3. Results

The leading *symptoms* resulting in the diagnosis of a pulmonary sequestration in these 14 patients were recurrent pneumonia in 10; five of them had a pulmonary abscess. Three patients had hemoptysis, and one had thoracic pain. The *diagnosis* including the localization of the feeding vessel was made by CT scan in all 14 patients. Three patients had an angiography, and one an angio-MRI additionally. Most patients were sent to our outpatient clinic with all these investigations previously done in other institutions.

There were 13 intralobar sequestrations. All of them were lower lobes, eight on the right and five on the left side. One patient had an extralobar sequestration on the left side posterior between diaphragm and lower lobe.

### 3.1. Surgery

All patients were operated fully thoracoscopically. One operation had to be converted to an open resection due to a bleeding from the aberrant artery caused by cautery with the hook electrode. There were eight thoracoscopic lobectomies (four left lower lobes and four right lower lobes), three atypical segmentectomies of the right lower lobe, and one atypical segmentectomy of the left lower lobe. In one patient with the leading symptom of hemoptysis and without evidence of inflammation in the history and preoperative CT scan, the feeding artery to the malformation was occluded during surgery with a stapling device (the knife was removed) (Multifire Endo GIA 30, 2.5 mm; Auto Suture, Tyco Healthcare). Finally, a lung biopsy was performed with a stapling device. The histological specimen was typical for a malformation with histological pulmonary hypertension with broadening of intima and media and thickened muscularis of the arteriolas. During the follow up of 4 years he had no further episodes of hemoptysis and was asymptomatic. In one case an extralobar sequestration was resected thoracoscopically.

The median operating time was 133 min (range 45–270 min). There was a considerable learning curve since this series includes the first thoracoscopic resection of pulmonary sequestration performed in our hospital. The median intraoperative blood loss, estimated by the anesthesiologist, was 200 ml (range 20–1200 ml) in thoracoscopic procedures. All patients had an intraoperative antibiotic prophylaxis.

### 3.2. Anatomy of vessels

In all intralobar sequestrations only one aberrant artery was found. In the extralobar sequestration two arteries could be identified. The aberrant artery originated in 10 cases from the descending thoracic aorta, in 3 cases from the abdominal aorta, and in 1 case from the right renal artery. The venous drainage was in the pulmonary vein in all cases except the extralobar sequestration, which drained into the hemiazygos vein.

### 3.3. Morbidity and mortality

There was no mortality. The postoperative complications included pneumonia in three, hemothorax in one, pneumothorax after removing of the chest tube in one and a superficial wound infection in one patient. All were treated conservatively. In the case of pneumothorax a chest tube was reinserted. The median postoperative hospitalization time was 7.5 days (range 3–13 days). In the long-term follow-up one patient suffered from intercostal neuralgias. Six months following the surgery he was free of symptoms. Another patient suffered from persistent chest pain. A pleurocutaneous fistula developed after 2 months due to an osteitis of the fifth rib and a pleural empyema. Six months following the decortication, resection of the rib, and excision of the fistula she was free of symptoms.

## 4. Discussion

It is generally accepted that resection is the treatment of choice in pulmonary sequestration [4]. Feasibility of VATS resection has been shown in case reports [5–22], or small series with two [23–25] or three cases [3,26]. To our knowledge this is the largest published report of thoracoscopic treatment of pulmonary sequestration.

The difficulty of the resection of a pulmonary sequestration is the identification of the aberrant artery. In our experience it is sometimes easy to identify the artery within the pulmonary ligament but most often there are inflammatory changes in the area of the sequestration due to recurrent infections and the artery is hidden in the scary tissue. These adhesions can be very dense, and scary tissue may mimic the artery. The identification of the aberrant artery or arteries should be done carefully at the beginning of the procedure. The localization of the vessel can be anticipated from the information of the preoperative imaging. In the early years of VATS lobectomy we occluded the artery centrally with a stapling device after removal of the knife and then we cut the artery peripherally with a second stapling device. With growing experience we recognized that this is not necessary and we meanwhile use only one stapling device even in very large vessels (Fig. 1). We did not observe a failure of a stapling device in any of our VATS lobectomies so far. One bleeding from the aberrant artery occurred during preparation of the aberrant artery. This complication could have been avoidable. In that case we clamped the artery with a lung forceps and converted to an open procedure. The intraoperative blood loss is relatively high in our series and is a consequence of the

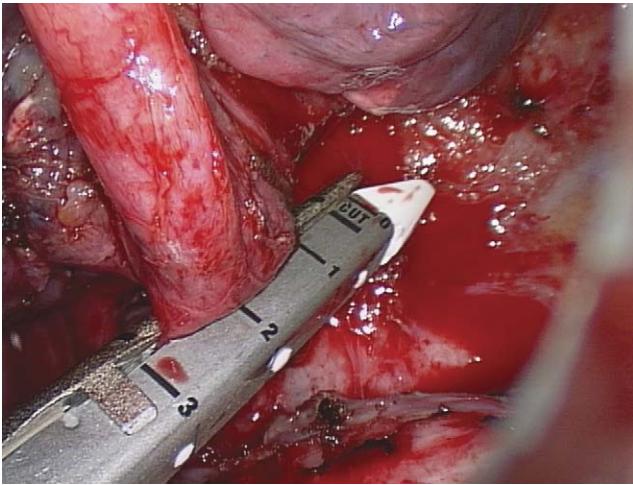


Fig. 1. Occlusion of the aberrant artery.

magnitude of the adhesions and inflammation especially in the cases with a pulmonary abscess. Surprisingly high is the rate of postoperative pneumonias in spite of using antibiotic prophylaxis. This may be due the fact that five patients had a pulmonary abscess and the antibiotic prophylaxis was not sufficient in these cases. As a consequence, we gain now sputum preoperatively to optimize the antibiotic prophylaxis and start an antibiotic therapy 5 days prior to the admission. Thus, we expect to reduce the hospital time.

In some cases patients were referred with an angiography performed additionally to the CT scan. In most cases this is not necessary because the feeding arteries of a sequestration can be identified very well on conventional images and 3D reconstructed images performed with a modern multislice CT scanner (Figs. 2 and 3).

In the same time period we performed five open resections of a pulmonary sequestration. Decision for a primary open approach was a planned anatomical segmentectomy in two cases or expected post-inflammatory adhesions in the early phase of VATS surgery.



Fig. 2. Aberrant artery (arrow) originating from the thoracic aorta.

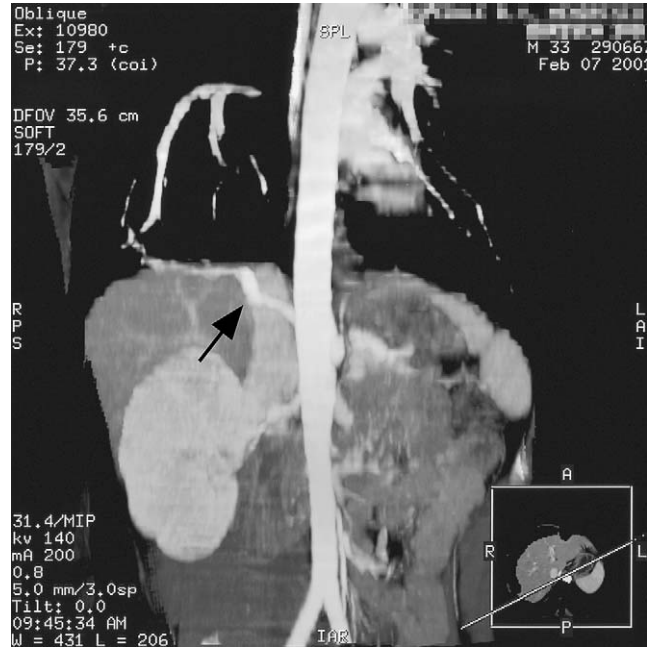


Fig. 3. Aberrant artery (arrow) originating from the abdominal aorta.

In conclusion, this study demonstrates that thoracoscopic treatment of pulmonary sequestration is feasible and can be performed safely. Therefore, the VATS approach is selected as a technique of choice in our group.

## References

- [1] Coman C, Stan A, Georgescu G, Dobrinov H, Dimitriu M. Present problems of intra- and extra-lobar pulmonary sequestration. *Poumon Coeur* 1973;29(2):211–20.
- [2] Savic B, Birtel FJ, Tholen W, Funke HD, Knoche R. Lung sequestration: report of seven cases and review of 540 published cases. *Thorax* 1979;34(1):96–101.
- [3] Weber A, Stammberger U, Inci I, Schmid RA, Dutly A, Weder W. Thoracoscopic lobectomy for benign disease—a single centre study on 64 cases. *Eur J Cardiothorac Surg* 2001;20(3):443–8.
- [4] La Quaglia MP. Congenital anomalies of the lung. In: Pearson GF, Cooper JD, Deslauriers J, Ginsberg RJ, Hiebert CA, Patterson GA, Urschel Jr HC, editors. *Thoracic Surgery*. 2nd ed., Philadelphia: Churchill Livingstone; 2002. p. 509–11.
- [5] Watine O, Mensier E, Delecluse P, Ribet M. Pulmonary sequestration treated by video-assisted thoracoscopic resection. *Eur J Cardiothorac Surg* 1994;8(3):155–6.
- [6] Mezzetti M, Dell'Agnola CA, Bedoni M, Cappelli R, Fumagalli F, Panigalli T. Video-assisted thoracoscopic resection of pulmonary sequestration in an infant. *Ann Thorac Surg* 1996;61(6):1836–7.
- [7] Arrabal Sanchez R, Benitez Domenech A, Pages Navarrete C, Fernandez de Rota Avecilla A, Fernandez Bermudez JL. Pulmonary sequestration: 2 cases (intralobar and extralobar) in surgically treated adults. *Arch Bronconeumol* 1998;34(1):45–7.
- [8] Nakamura H, Makihara K, Taniguchi Y, Ishiguro K, Ohgi S. Thoracoscopic surgery for intralobar pulmonary sequestration. *Ann Thorac Cardiovasc Surg* 1999;5(6):405–7.
- [9] della Porta M, Galli A, Rebuffat C. Videothoracoscopic approach in the treatment of intralobar pulmonary sequestration. Report of a case. *Chir Ital* 2000;52(3):299–302.
- [10] Gluer S, Scharf A, Ure BM. Thoracoscopic resection of extralobar sequestration in a neonate. *J Pediatr Surg* 2002;37(11):1629–31.
- [11] Wan IY, Lee TW, Sihoe AD, Ng CS, Yim AP. Video-assisted thoracic surgery lobectomy for pulmonary sequestration. *Ann Thorac Surg* 2002;73(2): 639–40.

- [12] Wang Z, Zhang Z, Yang C, Ren Y, Li B, Lin S. Clinical practice of video-assisted thoracoscopic surgery in children. *Zhonghua Wai Ke Za Zhi* 2002;40(6):401–3.
- [13] Klena JW, Danek SJ, Bostwick TK, Romero M, Johnson JA. Video-assisted thoracoscopic resection for intralobar pulmonary sequestration: single modality treatment with video-assisted thoracic surgery. *J Thorac Cardiovasc Surg* 2003;126(3):857–9.
- [14] Loscertales J, Congregado M, Arroyo A, Jimenez-Merchan R, Giron JC, Arenas C, Ayarra J. Treatment of pulmonary sequestration by video-assisted thoracic surgery (VATS). *Surg Endosc* 2003;17(8):1323.
- [15] Lupinski RW, Agasthian T, Lim CH, Chua YL. Extralobar pulmonary sequestration simulates posterior neurogenic tumor. *Ann Thorac Surg* 2004;77(6):2203–4.
- [16] Sakuma T, Sugita M, Sagawa M, Ishigaki M, Toga H. Video-assisted thoracoscopic wedge resection for pulmonary sequestration. *Ann Thorac Surg* 2004;78(5):1844–5.
- [17] Sato Y, Endo S, Saito N, Otani S, Hasegawa T, Sohara Y. A rare case of extralobar sequestration with hemoptysis. *J Thorac Cardiovasc Surg* 2004;128(5):778–9.
- [18] Tayama K, Eriguchi N, Tanaka A, Futamata Y, Harada H, Yoshida A, Matsunaga A. Video-assisted thoracic surgery lobectomy for extralobar pulmonary sequestration in a child: report of a case. *Surg Today* 2004;34(11):954–7.
- [19] Use T, Shimamoto H, Fukano T, Sumikawa K. Single lung ventilation in a pediatric patient using a Fogarty catheter with a hollow center. *Masui* 2004;53(1):69–71.
- [20] Hamanaka K, Nishimura H, Hosaka N. Extralobar pulmonary sequestration with aberrant lobulation of the lower lobe; report of a case. *Kyobu Geka* 2005;58(2):161–4.
- [21] Koizumi K, Haraguchi S, Hirata T, Hirai K, Mikami I, Kubokura H, Yamagishi S, Kawashima T, Yoshino N, Maeda M, Shimizu K. Thoracoscopic surgery in children. *J Nippon Med Sch* 2005;72(1):34–42.
- [22] Sato H, Watanabe A, Yamaguchi T, Harada N, Yamauchi A, Inoue S, Abe T. Pulmonary sequestration associated with asymptomatic aspergillosis. *Ann Thorac Cardiovasc Surg* 2005;11(1):41–3.
- [23] Albanese CT, Sydorak RM, Tsao K, Lee H. Thoracoscopic lobectomy for prenatally diagnosed lung lesions. *J Pediatr Surg* 2003;38(4):553–5.
- [24] Kaseda S, Aoki T, Shimizu K, Nakamura Y, Kiguchi H. Techniques for treating aberrant arteries during resection of pulmonary sequestration by video-assisted thoracic surgery: report of two cases. *Surg Today* 2003;33(1):52–4.
- [25] Tanaka T, Ueda K, Sakano H, Hayashi M, Li TS, Zempo N. Video-assisted thoracoscopic surgery for intralobar pulmonary sequestration. *Surgery* 2003;133(2):216–8.
- [26] Watanabe A, Osawa H, Watanabe T, Mawatari T, Ichimiya Y, Takahashi N, Abe T. Complications of major lung resections by video-assisted thoracoscopic surgery. *Kyobu Geka* 2003;56(11):943–8.

## Appendix A. Conference discussion

**Dr S. Mattioli (Bologna, Italy):** You have shown just regular CATscan. Don't you think that a TC angiogram or angio-TC scan or MRI could be helpful in identifying the aberrant vascularization just to render the thoracoscopic procedure even safer?

**Dr Kestenholz:** With the new 64 row multislice CT scanners, and three-dimensional reconstruction, it is quite easy to identify the aberrant artery. So we think it is mostly not necessary to perform angiography anymore.

**Dr G. Friedel (Gerlingen, Germany):** First, you have done four segmentectomies in the thoracoscopic group. I think that it could be a little bit difficult. What was the indication for segmentectomy? Were they all intralobar sequestrations?

**Dr Kestenholz:** These were all intralobar sequestrations and only a small part of the lobe had inflammatory changes. If severe inflammatory changes are present then we perform a lobectomy.

**Dr Friedel:** And the second question is, did you know in all cases before the operation whether these are sequestrations?

**Dr Kestenholz:** Yes.