



Uniportal thoracoscopic surgery for pulmonary arteriovenous malformations – report of technique and case series

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Background: Pulmonary arteriovenous malformations (PAVM) are abnormal direct connections between arteries and veins of the pulmonary circulatory system. In the majority of cases, they are of congenital origin and patients present with dyspnea, epistaxis or hemoptysis as the most common clinical symptoms. Before surgical treatment is considered, patients should be discussed interdisciplinary with angiologists and interventional radiologists regarding evaluation of embolotherapy. In case of unsuccessful embolotherapy, in case of complications (e.g., bleeding due to rupture of lesions), or in patients with contraindication for embolization such as untreatable contrast allergy, surgical resection of the pulmonary lesion is indicated. The former gold standard of open thoracotomy as treatment of PAVM has been mostly replaced by interventional embolization since the late 1980s. In recent years, less invasive approaches such as multi- or uniportal video-assisted thoracic surgeries (VATS) developed, have improved and have been shown to be safe and feasible treatment options, when surgery is necessary.

Methods: Between October 2015 and August 2019 five patients suffering from PAVM or its complications underwent a uniportal lung-sparing VATS resection at our department. Four out of the five patients have been successfully treated with a sublobar resection (segmentectomy of one or two lung segments). In another patient, an extended vascular malformation between the superior phrenic artery and the inferior pulmonary vein was approached via wedge resection of the affected lung in a first step and embolization of the subphrenic part in a second step as decided in an interdisciplinary setting before treatment.

Results: The median hospital stay was 4 days (range, 2–5 days) and, in all surgically treated patients, the postoperative course was uneventful. Only one of the five patients who suffered from hereditary hemorrhagic telangiectasia showed recurrence of the disease in the form of new vascular malformations in other lobes on CT scan 9 months after surgery, although so far asymptomatic. All other patients had an uneventful long-time course (median follow-up: 17 months; range, 5–52 months).

Conclusions: The uniportal VATS technique is one of the least invasive approaches in thoracic surgery: It allows not only for lung-sparing anatomical lung resections in the form of segmentectomies, but is also associated with very low morbidity and complication rates. Therefore, uniportal VATS is an excellent therapeutic option in selected cases in which primary interventional measures fail or complications of PAVM occur.

Keywords: Arteriovenous malformation; sublobar resection; uniportal lung resection; uniportal VATS technique; case series

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Introduction

Pulmonary arteriovenous malformations (PAVM) are vascular anomalies with a direct communication between pulmonary arteries and veins, bypassing the normal intervening capillary bed. By the majority, they are congenital, with dyspnea, epistaxis or hemoptysis as common clinical symptoms (1).

PAVM has firstly been described by Churton at autopsy in 1897 (2), and since then they have been given a great variety of names including pulmonary arteriovenous aneurysms or fistulas, hemangiomas or cavernous angiomas of the lung, pulmonary telangiectasias, or, as already mentioned, pulmonary arteriovenous malformations (1). The latter is the most commonly used term and will therefore be used throughout the following text.

The diagnosis of PAVM is made almost twice as frequently in women as compared to men. The estimated overall incidence is 38 per 100,000 persons (3). In up to 69%, a correlation is seen between PAVM and hereditary hemorrhagic telangiectasia (HHT), also known as Osler-Weber-Rendu syndrome (1). Conversely, PAVMs are present in up to 46% of patients with HHT (1,4,5). Although they can potentially affect any part of the lung, there is a predominance of the lower lobes (53–70%). In terms of localization, they can be found anywhere in the lung or spread over its surface (1,6,7).

PAVM can increase in number and size over time (7), therefore playing a role as differential diagnosis for evaluation of pulmonary nodules or hypoxemia and oxygen desaturation based on right-to-left shunting (8). About 70% of patients with PAVM are symptomatic (1,7). Bypassing the pulmonary capillary bed results in right-to-left shunt and potentially causes a variety of hypoxemic symptoms like exercise intolerance, dyspnea, cyanosis, digital clubbing, murmurs or bruits or even chest pain or cough (1,7).

The most frequent neurologic complications are transient ischemic attack, stroke or brain abscess and are most likely caused by paradoxical embolization based on the absence of a normal capillary bed, which normally acts as a filter for potential paradoxical embolisms (1,7,9). Rupture of PAVM can also lead to hemoptysis (endobronchial or intraparenchymal PAVM) or even hemothorax (e.g., superficial PAVM). Such bleedings can be life-threatening.

Diagnostic options are contrast enhanced CT scan of the chest or digital subtraction pulmonary angiography, measurement of shunt fraction (e.g., by 100% oxygen method) and contrast echocardiography (detection of right-

to-left shunts) (1,10,11). Avoiding ionizing radiation, contrast-enhanced MR perfusion imaging and MR angiography are other alternative diagnostic modalities (12,13).

The main treatment goal of PAVM includes prevention of neurologic complications (due to paradoxical embolization), progressive hypoxemia with its resulting complications (high-output cardiac failure) and hemorrhage (hemoptysis and/or hemothorax). Current guidelines recommend treatment of PAVM with a feeding artery diameter of 2 mm or larger (6,7,14-19). For smaller feeding arteries, the decision should be made on an individual basis (6,14-18), because these lesions also have a growth potential and pose a risk of enlargement as well as embolic complications at a later time (6,15,17-19).

The first 'successful' treatment of a patient with a PAVM was a pneumonectomy, performed by Shenstone in 1940 in a case, which was diagnosed by Hepburn and Dauphinée and published by them two years later in 1942 (20,21). Before Taylor published the first interventional treatment by using a device for embolization of PAVM in 1978 (22), conventional open thoracic surgery was the only known treatment option.

Being less invasive than conventional open thoracic surgery and easy to repeat, thenceforward the interventional approach with percutaneous embolization using for example coils replaced surgery in treatment of patients suffering from PAVM, thereby eventually being the most performed and recommended treatment for PAVM at present (6). All patients with PAVM should be evaluated by an interventional radiologist or angiologist for embolotherapy before being considered for surgery. Surgical resection of PAVM is indicated in patients who failed embolotherapy, develop serious bleeding complication despite embolotherapy, have intrapleural rupture of the PAVM or have untreatable contrast allergy and lesions not amenable to embolotherapy. Despite the fact that surgery has been mostly replaced by interventional options for treatment of PAVM in recent years, novel minimally invasive operative techniques advanced and offer a safe therapeutic option with low morbidity and mortality when surgery is necessary. Ongoing improvements in technique and instruments for video-assisted thoracoscopic surgery (VATS) have allowed lung surgery to become more minimally invasive with faster postoperative rehabilitation. Conventional three-port VATS is already well established for different types of lung resections. In recent years, uniportal VATS anatomical lung resection has become a new area of exploration in minimally invasive thoracic surgery. Uniportal VATS is



Figure 1 Uniportal approach with 4 cm incision and 24 French chest tube.

a less invasive approach that allows even major thoracic operations to be performed through a single small incision of some centimeters. Consistent reports confirmed different advantages of uniportal VATS, e.g., reduced surgical trauma, decreased postoperative pain, faster rehabilitation, and improved patient satisfaction with a less invasive approach than conventional VATS, but also with no significant differences in safety at the same time (23-25).

Methods

Our technique for uniportal VATS anatomical lung resection:

The procedure is performed under general anesthesia with a double-lumen endotracheal tube (DLT) or an endobronchial blocker. The patient is placed in a lateral decubitus position. The arms are flexed and positioned towards the head on two separate arm boards. The thoracic cavity is accessed by a 3–4 cm incision in the fourth or fifth intercostal space in the anterior to midaxillary line in a muscle-sparing technique (*Figure 1*). We routinely utilize a wound protector to maximize wound exposure, avoid soiling of the camera and protect the wound from contamination. No rib spreader is used. To explore the pleural cavity, a

5 mm 30° angled endoscope is used.

Vascular dissection can be performed analogous to any thoracoscopic anatomical resection. The targeted branches of the pulmonary artery are dissected and cut with endoscopic stapling devices prior to the vein, avoiding pressure dependent rupture of the pulmonary arteriovenous malformation. After dividing the artery, vein and bronchus, if deemed necessary, a systemic injection of indocyanine green (ICG) can be used in combination with near-infrared imaging to identify and mark the intersegmental planes.

Ethical approval was not required because of the descriptive manner of this study with limited participants (not more than 5). The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). Written informed consent was obtained from all patients for publication of this manuscript including accompanying images.

Patients

From October 2015 to August 2019, we performed uniportal VATS lung resections for PAVM in five patients. Four of these five patients underwent a lung-sparing uniportal VATS segmentectomy (3 single segmentectomies and one lingula resection) and in one patient, a uniportal wedge resection combined with embolotherapy was scheduled and performed for a complicated and extensive arteriovenous malformation between the superior phrenic artery and the right inferior pulmonary vein. Patient characteristics are summarized in *Table 1*.

Three of five patients were female and in four patients PAVM was located in the lower lobe. One had recurrent disease with two PAVM in both lower lobes treated already in the past (Patient E). Two patients were diagnosed with hereditary hemorrhagic telangiectasia (C, E). Multiple lesions were only found in patient E, which have been previously treated by embolization in multiple sessions years before actual surgical treatment. The malformations of patients A, B, C and E could be accurately localized in one single lung segment or between two adjacent segments (lingula), well accessible by the uniportal minimal-invasive approach. Diagnostics included pulmonary function tests with respiratory measurement with exercise electrocardiogram, chest X-ray, computed tomography angiography (CTA) or magnetic resonance angiography (MRA) and evaluation for hereditary hemorrhagic telangiectasia (selected radiographic or angiographic findings are illustrated in *Figure 2*).

Clinical manifestations ranged from epistaxis without

Table 1 PAVM patient characteristics

Patient	Age (years)	Sex	Number of PAVM	Localization	HHT Symptoms	Embolotherapy	Indication for surgery
A	20	F	1	RS8	No Dyspnea on exertion	Declined	Interdisciplinary discussion: patient prefers surgery
B	57	F	1	LS6	No Desaturation on exertion, stroke	–	Referral from cardiologist
C	15	M	1	RS6	Yes Epistaxis	–	Referral from otolaryngologist
D	29	F	1	Superior phrenic artery and right inferior pulmonary vein	No Dyspnea on exertion	Unsuccessful, embolotherapy, decision for surgery combined with ethanol embolization afterwards	Interdisciplinary decision for combination after unsuccessful embolization
E	37	M	Multiple AVM	Lingula, RUL, both LL, brain	Yes Epistaxis, chest pain, dyspnea, repetitive hemothorax	Coiling of PAVM in lingula plus preventive coiling of left bronchial artery during first hemothorax	Massive hemothorax

PAVM, pulmonary arteriovenous malformations; AVM, arteriovenous malformation; RS8, right segment 8; LS6, left segment 6; RS6, right segment 6; RUL, right upper lobe; LL, lower lobe.

any respiratory symptoms (C) to hypoxemic symptoms and one stroke as the only neurologic complication in patient B. Patients A, B, D and E presented with dyspnea or oxygen desaturation on exertion. Additionally, patient E previously showed multiple clinical manifestations including epistaxis and chest pain due to hemothorax, following coiling of one bronchial artery and embolotherapy of the lingular PAVM with vascular plugs. Eight months later repetitive hemothorax occurred, presenting with ipsilateral chest pain, dyspnea and oxygen desaturation.

Indication for surgery was made based on number, size, distribution and localization of the malformations in an interdisciplinary setting.

Results

Five patients with pulmonary arteriovenous malformations were treated by uniportal minimally invasive thoracic surgery in the last four years. Performed resections, postoperative parameters and outcome including length of hospital stay, chest tube duration and follow-up time as well as complications and their management are listed in *Table 2*. Regarding the four solitary lesions which were completely located in the lung parenchyma, (A, B, C, E) anatomical resection of the involved single segment or both lingular segments in E was performed using the uniportal VATS approach reported before. In patient E, one additional and

smaller ipsilateral PAVM, which has been coiled previously was left untreated because of stable size and missing signs of revascularization. The vascular confluence between the superior phrenic artery and the inferior pulmonary vein in patient D was locally excised right above the diaphragm and treated by embolization afterwards in a second step as planned preoperatively based on multidisciplinary clinical case discussion after unsuccessful embolization in a first step.

The median postoperative day (POD) for chest tube removal was two (range: 1 to 3) and patients were discharged between POD 2 and POD 5 (median: 4). In patient E with hemothorax as complication of PAVM, the chest tube was removed on POD 3 with discharge on POD 5.

There were no complications regarding PAVM during follow-up in all patients except in Patient E. In this patient with hereditary hemorrhagic telangiectasia, there were some new malformations found in other lobes than the operated one in CT scan follow-up 9 months after operation, without any complications or symptoms so far. Patient C showed a postoperative airspace slightly bigger than expected after segmentectomy with some subcutaneous emphysema after chest tube removal, which was successfully treated conservatively in an otherwise asymptomatic patient. Furthermore, the patient (D) who has undergone local excision of a malformation between the superior phrenic artery and the inferior pulmonary vein showed an elevated ipsilateral hemidiaphragm in the chest X-ray after

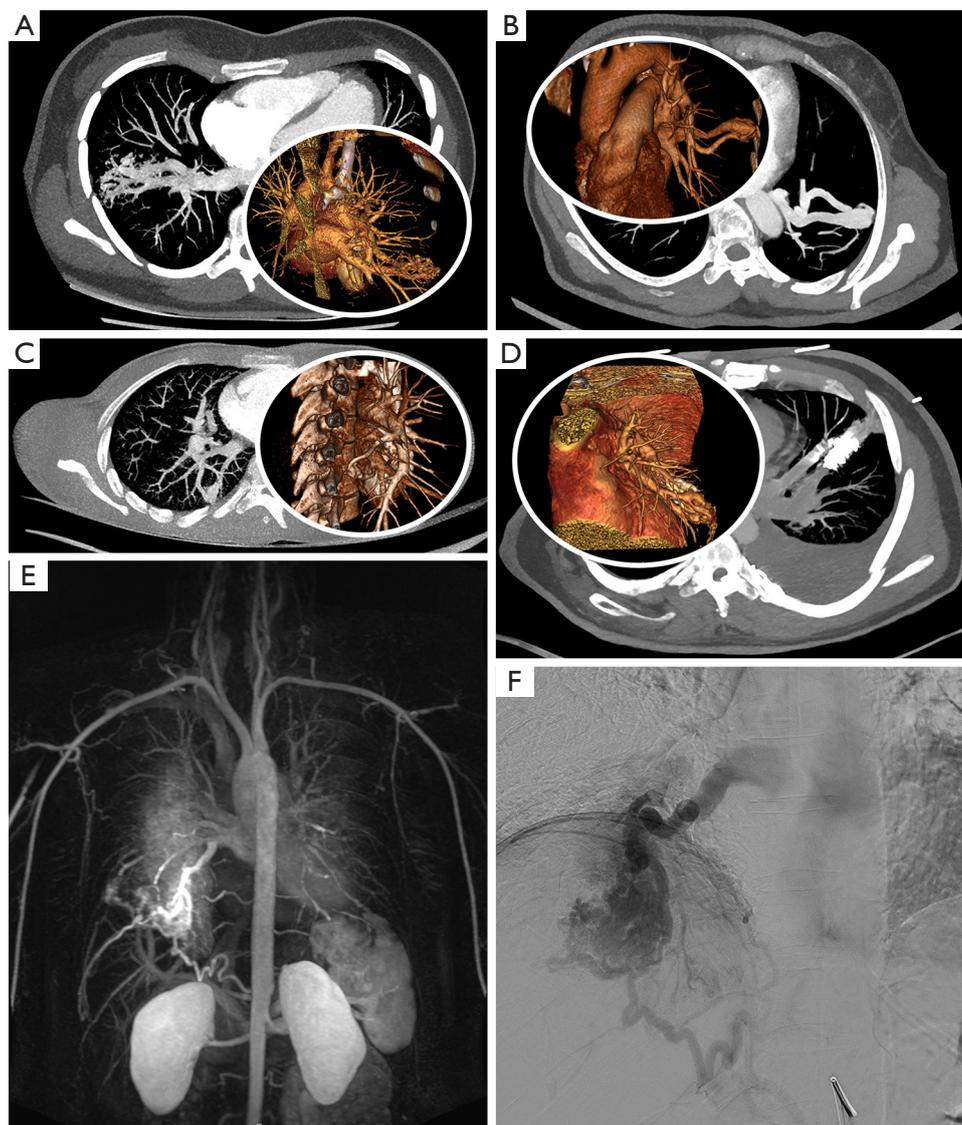


Figure 2 Radiographic findings: (A) pulmonary arteriovenous malformations (PAVM) in segment 8 of the right lower lobe; (B) PAVM in segment 6 of the left lower lobe; (C) PAVM in segment 6 of the right lower lobe; (D) PAVM in lingula with left hemothorax; (E) 3D-reconstruction of angiography of a PAVM of the right superior phrenic artery and the right inferior pulmonary vein; (F) digital subtraction angiography of same PAVM as described in (E).

tube removal as an incidental finding without any clinical complaints. No further diagnostics were made, since the patient was free of symptoms. There was a complete recovery in follow-up check-up. Overall follow-up period was up to 52 months postoperatively.

Discussion

Conventional open thoracic surgery has been replaced as

preferred therapeutic option for PAVM by effective and less invasive interventional embolization techniques in the past, but in recent years thoracoscopic pulmonary surgery has also very much advanced (26). Technical feasibility, safety and advantages of VATS (27), especially for major anatomical lung resections, has been demonstrated in different meta-analyses (28,29), systematic reviews (30,31) and in prospective multi-institutional studies (32). Furthermore, even less invasive techniques have been

Table 2 Treatment and outcome of PAVM patients

Patient	Operation	Drain removal (POD)	Discharged (POD)	Postoperative complications	Complication management	Follow-up
A	uVATS anatomical RS8 resection	1	2	None	–	Free of complaints at 52 months
B	uVATS anatomical LS6 resection	1	5	None	–	Free of respiratory complaints at 31 months, persistent amnesic aphasia since preoperative stroke
C	uVATS anatomical RS6 resection	2	4	Pneumothorax, subcutaneous emphysema	Conservative	Regressive airspace and emphysema, free of complaints at 5 months
D	uVATS wedge resection of vascular confluence in RLL & supradiaphragmatic dissection	2	2	None	–	Free of complaints at 8 months
E	uVATS anatomical lingula resection, evacuation of hematoma on the left	3	5	None	–	Increased numbers of AVM, no chest pain or dyspnea at 17 months

PAVM, pulmonary arteriovenous malformations; AVM, arteriovenous malformation; POD, postoperative day; uVATS, uniportal video-assisted thoracic surgery; RS8, right segment 8; LS6, left segment 6; RS6, right segment 6; RLL, right lower lobe.

developed in the last three decades. The technique of using a single small incision for VATS has revolutionized the way thoracic surgeons treat pulmonary diseases today (23,26). With the involvement of only one intercostal space and without using any rib spreading, single-incision video-assisted thoracoscopic anatomic resections of almost every extent can be performed without compromising safety compared to the traditional multi-portal approach. Furthermore, the uniportal VATS approach can potentially improve perioperative outcomes such as reduction of overall rate of complications, faster recovery with shorter length of hospital stay and shorter chest tube duration (24,25).

Although current guidelines (6,14-16,19) are still prioritizing embolotherapy, these recommendations and results originate from retrospective observational non-controlled case series and prospective cohort studies (6). At present, no direct comparison has been made between embolotherapy and minimal invasive surgical techniques such as any type of VATS in treatment of patients with PAVM.

Although there is no high level of evidence for surgery in treatment of PAVM, retrospective case reports and case series have shown good results (8,33-37).

In the literature mentioned above, indications for resection of the PAVM using a minimal invasive surgical approach are rather vague and no clear selection criteria have been formed so far. Indication for pulmonary resection as treatment of PAVM should be based on number, size, distribution and localization of the malformation and

furthermore, should be an individual decision made by a multidisciplinary board. Moreover, patients' preference and general health conditions may be respected as well.

The extent of surgical resection is determined by location and size of the lesion. Extra-anatomical wedge resection or anatomical resections such as segmentectomy, lobectomy or even pneumonectomy are possible types of resection (6). Lung transplantation is reported but not recommended (38,39). Multiple small PAVM can occur, persist or even new ones may be formed over time, especially in individuals with HHT, hence, neither embolization of detectable lesions nor surgery will be able to cure these patients and prevent them from developing new malformations (6). In these patients embolization might be considered over surgery due to avoidance of tissue loss after potential repetitive excision or resection of all detected lesions (6) (Appendix 1).

It is important to emphasize that the indication for treatment of individuals with PAVM should be based on multidisciplinary board recommendations and each case should be discussed separately to provide the most beneficial treatment option for every patient.

If no contraindications are present, interventional embolization approaches or combinations with embolotherapy and surgery as shown in our patient D can be considered. In case of unsuccessful embolotherapy or if complications occurred, surgery offers a safe therapeutic alternative.

Thanks to huge changes in thoracic surgery and

development of minimal invasive approaches, patients can benefit from these new surgical techniques and VATS might be even considered as equal therapy modality in selected cases. However, considering the limited evidence in the form of only retrospective studies and small case series, further studies are needed to investigate this issue.

Conclusions

In all of the presented cases, uniportal VATS was performed effectively. In selected cases of patients with PAVM with easily accessible lesions, in cases of unsuccessful embolization, or in complications, minimal invasive thoracic surgery approaches such as the uniportal VATS technique are safe and feasible therapeutic options, which are well tolerated especially in combination with lung sparing resections.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://cts.amegroups.com/article/view/10.21037/cts.2020.04.01/coif>). GJK serves as an unpaid editorial board member of *Current Challenges in Thoracic Surgery* from September 2019 to August 2021; PD serves as an unpaid editorial board member of *Current Challenges in Thoracic Surgery* from December 2019 to November 2021. The other authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Ethical approval was not required because of the descriptive manner of this study with limited participants (not more than 5). Written informed consent was obtained from all patients for publication of this manuscript and any accompanying images. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013).

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Supplementary: Discussion



Dr. Hon Chi Suen (Hong Kong, China)

“I would like to congratulate the authors for successfully using uniportal thoracoscopic surgery to treat selected patients with pulmonary arteriovenous malformation. It is definitely a logical step to apply the most minimally invasive thoracoscopic technique in this situation. The fact that we can now perform precise uniportal thoracoscopic segmentectomy makes this approach appealing. I have a provoking question:

Can we make the surgical approach even more minimally invasive? If we apply the principle of embolization (that is to block the feeding artery only), can we just precisely locate the feeding artery and then staple and divide it without resecting any lung? That would be even more minimally invasive! Would this approach result in pulmonary infarction? I am curious about the authors’ thought on this.”



Dr. Patrick Dorn (Bern, Switzerland)

“Thank you very much, Dr Suen, for this question. Indeed, your idea is an interesting approach we have already thought about.

Technically, dividing only the feeding artery is feasible in most cases of simple PAVMs. Of course, it is going to be more challenging in complex cases, where for example two or more feeding arteries are supplying the malformation, which is sometimes the case based on our experience and also according to the literature we mentioned. We would expect pleuritic chest pain as most common complication after selective division of the feeding vessel only, what is already shown after interventional embolotherapy. This reaction seems to be successfully treated with pain killers in most cases.

Thanks to the systemic lung supply by bronchial arteries, lung infarction and consecutive infection are rarely seen. Nevertheless, we think that the minimally invasive surgical treatment of PAVM has the potential to assert itself as an attractive option in complicated situations or unsuccessful embolotherapy.

Additionally, we assume that the risk for recurrent disease is lower after resecting the corresponding lung parenchyma of the whole PAVM including feeding and draining vessels compared to dividing the feeding vessel only, but there is not yet any evidence about that in the literature.”