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Aberrant artery embolization prior to pulmonary sequestration surgery: A case series report

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ABSTRACT

Objectives: Pulmonary sequestration is a rare congenital malformation, and part of its treatment requires the removal of the aberrant artery by surgical means.

Materials and methods: Five patients treated at Military Hospital 103 - Department of Thoracic Disease were diagnosed with PS via CT scan, MS-CT, and DSA, and histopathological data were evaluated retrospectively between January and December 2019.

Results: In all patients, surgery is the preferred option, with two cases of video-assisted thoracoscopic surgery (one lobectomy and one wedge resection), and three cases of hybrid video-assisted thoracoscopic surgery (adhesive inflammation was observed, the bronchus is challenging to reveal, to resect, and tend to bleed when resecting).

The average length of stay following surgery is 11.6 ± 8.1 days. The mean duration of postoperative follow-up is 13.8 ± 3.3 months, all patients had a good quality of life, and no respiratory problems such as hemoptysis or pneumonia were detected.

Conclusion: The excellent outcomes obtained in all patients in our study during the follow-up period (13.8 ± 3.3 months) established the appropriate indication and treatment. However, these are preliminary findings; a longer study period with a larger sample size is required to draw more valid conclusions.

1. Introduction

Pulmonary sequestration (PS), alternatively referred to as the accessory lung, is a cystic or solid mass of nonfunctioning primitive segmental lung tissue that is isolated from the tracheobronchial tree and has an abnormal systemic blood supply.

It is categorized anatomically into two types: intralobar sequestration (ILS) and extralobar sequestration (ELS), depending on the aberrant segmental lung tissue's relationship to the aberrant segmental lung tissue pleura. Sequestration within the lobe is more common.

PS is a rather uncommon congenital lung abnormality. Between January 1st, 2005 and December 31st, 2015, at the National Tuberculosis and Lung Diseases Research Institute in Warsaw, Poland, only 25 cases of PS were diagnosed out of 110.000 hospitalized patients. In general, the incidence of PS is estimated to be 6.4% [1–3]. In Vietnam, studies on PS are still uncommon. We discussed several cases of PS sequestration to demonstrate the utility of angiography in diagnosing and embolizing the aberrant artery prior to performing pulmonary lobectomy to treat this condition.

2. Materials and methods

Five patients treated at Military Hospital 103 - Department of Thoracic Disease were diagnosed with PS via CT scan, MS-CT, and DSA, and histopathological data were evaluated retrospectively between January and December 2019. We collected and analyzed clinical and subclinical data (chest computed tomography, MSCT, and angiography).

Clinical examinations and CT scans are performed at 3-month and 6-

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Table 1
Patients characteristics, causes of admission, and prior diagnoses.

Patients	Age	Gender	Causes of admission	Prior diagnoses
1	25	Male	Fever, cloudy sputum cough	Lung abscesses
2	52	Female	Hemoptysis	Suspected Lung cancer
3	68	Male	Fever, cloudy sputum cough	Pneumonia
4	52	Male	Cloudy sputum cough	Pneumonia
5	55	Female	Hemoptysis	Suspected Lung cancer

Table 2
Chest CT and MSCT results.

Patients	Size of mass (mm)	Sublobar location	Aberrant artery defined by	
			CT	MSCT
1	65 × 90 × 96	9. 10 (L)	Yes	Yes
2	46 × 75 × 76	10 (L)	No	Yes
3	39 × 52 × 66	10 (L)	Yes	Yes
4	54 × 37 × 32	7 (L)	No	Yes
5	51 × 40 × 46	7 (L)	Yes	Yes

month intervals following surgery. We finalize the follow-up on 01 June 2020 (Table 1 and 2).

In all cases, angiography was performed (Fig. 1). The angiography results indicate that there are no variations in feeding artery findings between the angiography and MSCT outcomes. Embolization is then conducted to prepare for the operation, with PVC particles and spongel employed as embolization materials.

This report can be used for educational purposes and clinical practice. This case report was drafted and submitted according to the PROCESS 2020 Guidelines [4].

3. Results

Five patients were diagnosed and operated on in our study at Military Hospital 103's Department of Thoracic Surgery. In all cases, we observe intralobar PS in the left lung inferior lobe. Three male patients and two female patients participated in our investigation. Patients range in age from 26 to 70 years. All individuals presented with intralobar PS in the inferior lobe of the left lung.

Pneumonia-related symptoms (fever, cloudy sputum) were observed in 03 cases and hemoptysis in 02 cases.

In all five patients, chest CT (5 mm slice thickness) revealed mass lesions located in the left lung inferior lobar basal sublobar (sublobar 7, 9, 10), ranging in size from 39x45x45 mm to 65x90x96 mm. In 2 cases, we can define the aberrant artery branch from the thoracic aorta. MSCT results defined the aberrant artery in all cases, with one being a branch of the abdominal aorta, one being a celiac artery branch, and three being thoracic aorta branches. Each case drains via the inferior lung vein (Fig. 2).

In all patients, surgery is the preferred option, with two cases of video-assisted thoracoscopic surgery (one lobectomy and one wedge resection), and three cases of hybrid video-assisted thoracoscopic surgery (adhesive inflammation was observed, the bronchus is challenging to reveal, to resect, and tend to bleed when resecting). All patients was performed by Dr. Vu Anh Hai who was expert on thoracoscopic surgery. All patients was followed standard protocol of thoracoscopic surgery that approved by Military Hospital 103 to ensure quality control.

We always conduct dissection and resection of the aberrant artery first by cutting the pulmonary ligament, and then we dissect progressively upward until the aberrant artery is revealed and then cut using the stapler. During dissection, we observed that the aberrant artery laid under the inferior pulmonary vein, and one case with the aberrant artery laid above the inferior pulmonary vein.

We saw one case of a prolonged air leak that was successfully treated with chest tube re-intubation and pleurodesis using Povidone 4%. The average length of stay following surgery is 11.6 ± 8.1 days (shortest is six days and longest is 26 days). The mean duration of postoperative follow-up is 13.8 ± 3.3 months (range: 10 to 17 months), all patients had a good quality of life, and no respiratory problems such as hemoptysis or pneumonia were detected.

4. Discussion

According to Polaczek M. et al. (2017) [5] the mean age of PS patients receiving surgical treatment was 38.24 ± 14.93, the youngest was 15, the oldest was 67 years old. The report of Sun X. et al. (2015) [6] also gives similar results, with the mean age of patients being 36.6 ± 11.8. From 1998 to 2008, Wei Y et al. (2011) published a review article with a large population of 2625 cases. The author mentioned that the age of patients with PS detection ranges from 1 month to 77 years. According to the author, extralobar PS patients are more likely to be detected at an older age than intralobar PS patients (38 9 vs. 20 8). To explain this explanation, the author hypothesizes that inflammation occurs as a result of the anatomical structure of the bronchopulmonary intralobar PS with communication to the bronchial tree. Therefore, patients often

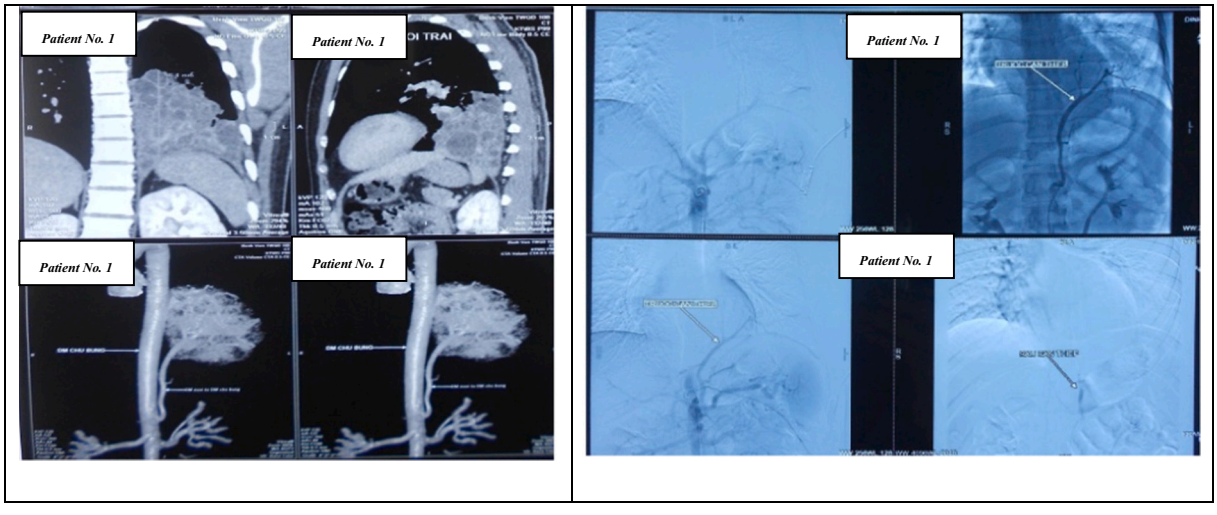

Fig. 1. Aberrant artery of the PS on MSCT and angiography.

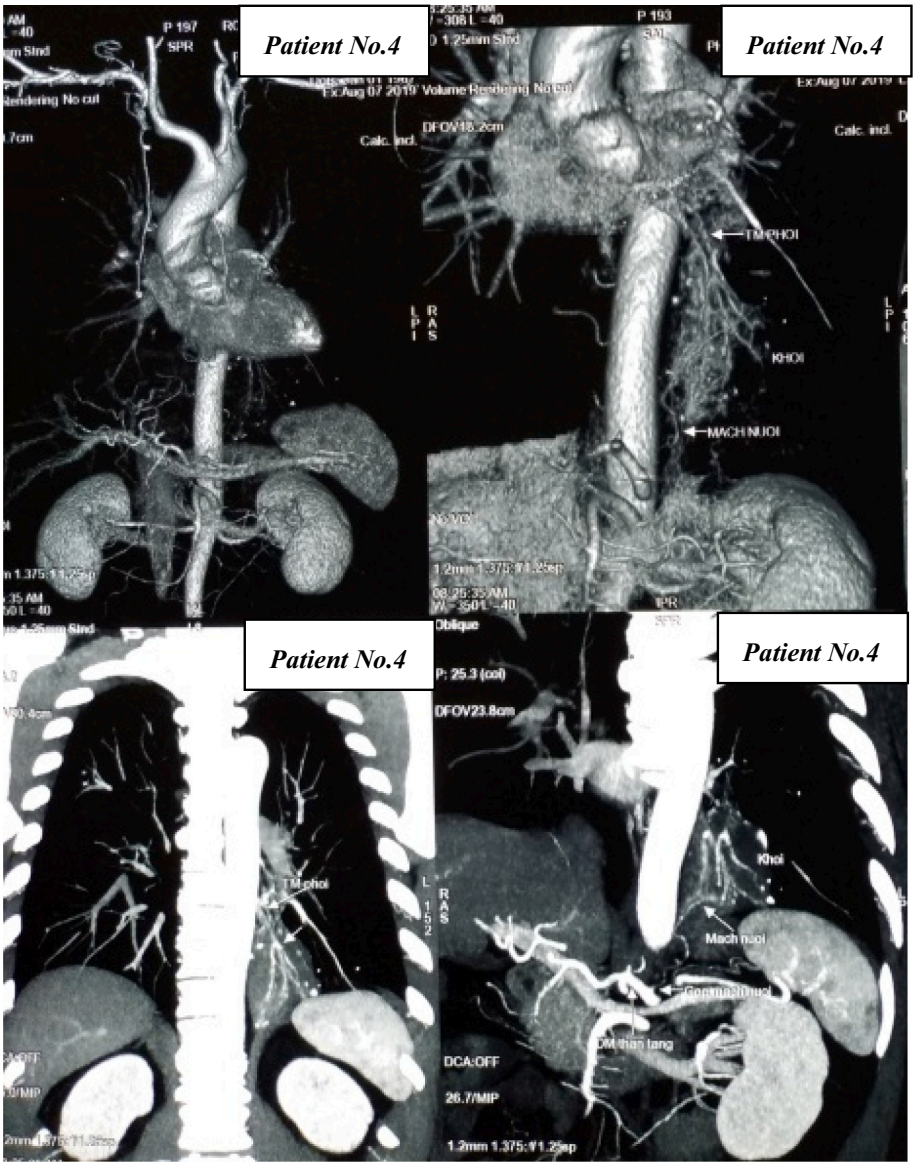

Fig. 2. PS and its aberrant artery.
**Sources*: *Patient No.4*
MSCT results defined the aberrant artery as a branch from the celiac artery.

have early respiratory symptoms and are examined and detected early [7]. In our study, study patients aged from 26 to 70, mean age is 52.2 ± 16.1.

The typical clinical symptoms in PS patients are cough and sputum production (67.76%), fever (37.9%), hemoptysis (27.7%) [7] In many cases, symptoms were recurring, persistent with manifestations of pulmonary abscesses, and pus production that responded poorly to antibiotics [6].

CT has limited value in diagnosis. According to Sun X., et al. (2015), PS diagnosis rate by CT with contrast injection only reached 37.5% [6]. We found 2 out of 5 instances (40%) with anomalous, aberrant arteries providing blood to the lesion using contrast injection CT scan, assisting in guiding the PS diagnosis. MSCT scans are quite valuable in diagnosis. It accurately identifies the arterial branch of the system that provides blood to the isolated lungs, and so is recommended as the first line of defense in diagnosis [8]. Our study's findings also lend weight to this assertion. MSCT scans identify all individuals with abnormal arteries feeding blood and drainage veins, providing a conclusive diagnosis.

Embolization is a minimally invasive procedure, being applied widely in the treatment of PS, especially in children [9] with the complication of hemoptysis [3,10]. However, its outcome and effectiveness require more assessment. When administered alone, embolization has a recurrence rate of between 25% and 47% [11]. In certain situations, the patient underwent embolization three times but was ultimately unsuccessful; the patient developed recurrent hemoptysis and was effectively treated by endoscopic pulmonary lobectomy.

Surgical treatment demonstrates its advantage by completely resolving the lesions, the mass. It may be performed openly or with video-assisted thoracoscopic surgery (VATS) [6]. VATS is a minimally invasive procedure that provides aesthetics and enables patients to recuperate rapidly following the procedure. Despite numerous reports of feasibility, it is technically difficult, with a high risk of complications from excessive bleeding during surgery due to pleural thickness, problems during surgery, and vivisect of the aberrant artery [5]. Combining angiography, pre-operative aberrant artery embolization, and PS removal surgery has a number of advantages: it has been shown to be useful in emergency hemoptysis situations and in elevating the patient's status for procedural pulmonary lobectomy [10], initializing embolizing

also helps the surgeons to define the aberrant artery easier in case the pulmonary ligament has become severely inflamed; due to the rigid nature of the embolizing material within the blood vessel. Additionally, it decreases the likelihood of problems such as bleeding during surgery due to recurring chronic inflammation. In general, indicating angiography aids in the definitive identification and preoperative therapy of the aberrant artery, while minimizing postoperative bleeding.

We perform intravascular intervention with two objectives in mind: definitive diagnosis and embolization in preparation for surgery. As a result of the results, hemoptysis patients who received pre-operative embolization (2/5 cases) had a considerable improvement in their condition and were no longer suffering from hemoptysis. Then we did lobectomy: the surgeons are relaxed and confident throughout the procedure due to the well-defined aberrant artery.

5. Conclusion

The excellent outcomes obtained in all patients in our study during the follow-up period (13.8 $\pm$ 3.3 months) showed the utility of the aberrant artery embolization prior to pulmonary sequestration surgery. However, these are preliminary findings; a longer study period with a larger sample size is required to draw more valid conclusions.

Data sharing statement

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Ethical approval

The Ethics Committee of Vietnam Military Medical University approved the study and authorized its conduct and follow-up. The study was in line with the Declaration of Helsinki. Individual patient consent for inclusion in the study was obtained. Before operation, written informed consent was provided to all participants after a thorough explanation of the purpose of this study. Patients had the right to discontinue at any time during the study.

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Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

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Registration of research studies

This is a case series report that does not require a research registry.

Guarantor

Vu Anh Hai.

CRediT authorship contribution statement

All authors made substantial contributions to conceptualization and design, data acquisition, data analysis and interpretation, took part in drafting of the initial manuscript and revising it critically, gave final approval of the version to be published, agreed to submit to the current journal, and agreed to be accountable for all aspects of the work.

Declaration of competing interest

The authors declare that they have no conflicts of interest for this work.

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Non-applicable.

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