Türk Göğüs Kalp Damar Cerrahisi Dergisi 2017;25(1):96-102
http://dx.doi.org/doi: 10.5606/tgkdc.dergisi.2017.13104

Original Article / Özgün Makale

Surgical approach in pulmonary sequestrations: An institutional experience with 32 cases

Pulmoner sekestrasyonlarda cerrahi yaklaşım: 32 olgu ile kurumsal deneyim

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ABSTRACT

Background: This study aims to present our experiences on surgical treatment of pulmonary sequestrations.

Methods: A total of 32 patients (16 males, 16 females; mean age 32.7±13.4 years; range 4 to 61 years) who were performed surgical resection for pulmonary sequestration between January 1987 and December 2015 were reviewed. Patients were evaluated according to demographics, symptoms, diagnostic methods, type, localization and vascular characteristics of pulmonary sequestration, type of resection, complications, and outcomes.

Results: Of the patients, intralobar sequestration was detected in 87.5% and extralobar sequestration in 12.5%. Intralobar sequestration was located in the lower lobe in 89.3% and in the upper lobe in 10.7% of the patients. All extralobar sequestrations were located in the left hemithorax; 75% was located at the base of the hemithorax and 25% was located at the apex. Of the patients, lobectomy was performed in 53.1%, wedge resection in 25%, mass excision in 12.5%, and segmentectomy in 9.4%. Postoperative complications occurred in 18.8% of the patients as prolonged air leak in three patients, wound infection in two patients, and empyema in one patient. No mortality was observed. Median follow-up period was 42 months (range 3 to 105 months) and we detected that patients remained asymptomatic.

Conclusion: Although pulmonary sequestration has a benign course, it should be removed owing to its severe complications even if patients are asymptomatic. Computed tomographic angiography is a good diagnostic tool for pulmonary sequestrations.

Keywords: Computed tomographic angiography; pulmonary sequestration; surgery.

ÖZ

Amaç: Bu çalışmada, pulmoner sekestrasyonların cerrahi tedavisi hakkında deneyimlerimiz sunuldu.

Çalışma planı: Pulmoner sekestrasyon nedeniyle Ocak 1987 - Aralık 2015 tarihleri arasında cerrahi rezeksiyon uygulanan toplam 32 hasta (16 erkek, 16 kadın; ort. yaş 32.7±13.4 yıl; dağılım 4-61 yıl) incelendi. Hastalar demografik özellikleri, semptomlar, tanı yöntemleri, pulmoner sekestrasyonun tipi, yerleşim yeri ve vasküler özellikleri, rezeksiyon tipi, komplikasyonlar ve sonuçlara göre değerlendirildi.

Bulgular: Hastaların %87.5'inde intralober, %12.5'inde ekstralober sekestrasyon saptandı. Intralober sekestrasyon hastaların %89.3'ünde alt lob, %10.7'sinde üst lob yerleşimli idi. Bütün ekstralober sekestrasyonlar sol hemitoraksta olmak üzere; %75'i hemitoraks tabanı, %25'i ise apeks yerleşimli idi. Hastaların %53.1'ine lobektomi, %25'ine kama rezeksiyon, %12.5'ine kütle eksizyonu, %9.4'üne segmentektomi uygulandı. Ameliyat sonrası komplikasyonlar üç hastada uzamış hava kaçağı, iki hastada yara yeri enfeksiyonu ve bir hastada ampiyem olmak üzere hastaların %18.8'inde gelişti. Mortalite izlenmedi. Ortanca takip süresi 42 ay (dağılım 3-105 ay) idi ve hastaların asemptomatik kaldığı saptandı.

Sonuç: Pulmoner sekestrasyon benign seyirli olmasına rağmen ciddi komplikasyonları nedeni ile hastalar asemptomatik olsa da çıkarılmalıdır. Bilgisayarlı tomografik anjiyografi pulmoner sekestrasyonlara yaklaşımda cerraha yol gösteren iyi bir tanı aracıdır.

Anahtar sözcükler: Bilgisayarlı tomografik anjiyografi; pulmoner sekestrasyon; cerrahi.

Available online at
www.tgkdc.dergisi.org
doi: 10.5606/tgkdc.dergisi.2017.13104
QR (Quick Response) Code

Received: February 08, 2016 *Accepted:* February 25, 2016

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Pulmonary sequestration is a non-functioning lung tissue which has no connection with the normal tracheobronchial tree and receives its blood supply from the systemic circulation. This rare abnormality constitutes 0.15% to 6.4% of all congenital pulmonary malformations.[1] Pulmonary sequestrations are recognized as either intralobar sequestration (ILS) or extralobar sequestration (ELS), based on whether they are contained within the normal pulmonary parenchyma and have their own visceral pleura.[2] In this article, we aimed to present our experiences on surgical treatment of pulmonary sequestrations.

PATIENTS AND METHODS

A total of 32 patients (16 males, 16 females; mean age 32.7±13.4 years; range 4 to 61 years) with pulmonary sequestration who underwent operation in the Department of Thoracic Surgery in Ankara University Faculty of Medicine between January 1987 and December 2015 were evaluated according to gender, age, symptoms, type of sequestration and localization, arterial blood supply, venous drainage, type of resection, complications, and outcomes. All patients were preoperatively evaluated with routine blood and pulmonary function tests, chest radiography, and bronchoscopy. Computed tomography (CT) was performed for all except nine patients, as it only came into use in our institution in 1993. In addition, computed tomographic angiography (CTA), digital subtraction angiography (DSA), and magnetic resonance angiography (MRA) were used for further confirmation in certain patients as advanced imaging methods. Posterolateral thoracotomy was performed in all patients. The study protocol was approved by the Medical Faculty of Ankara University Faculty Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki.

RESULTS

Cough was the most common symptom in 21 patients, whilst seven (21.9%) patients (all ELS patients and only three ILS patients) were asymptomatic. Chest radiography was the only radiological method performed in nine patients before 1993. Thereafter, CT was routinely used as an initial diagnostic method. Computed tomography could provide the vascular anatomy in only four (17.4%) of 23 patients. The lesion was further evaluated with DSA in five patients, with CTA in four patients, and with MRA in two patients (Table 1) (Figures 1, 2). Pulmonary sequestration was localized on the left side in 23 patients (71.9%) and on the right side in nine patients (28.1%). There were 28 (87.5%) ILS and four (12.5%) ELS patients. The ILS was located in the left lower lobe in 17 patients (60.7%), the right lower lobe in eight patients (28.6%), the left upper lobe in two patients (7.1%), and the right upper lobe in one patient (3.6%). All ELSs were located in the left hemithorax. Whilst three of four ELSs were located at the base of the hemithorax above the diaphragm, ELS was located at the apex in the remaining one patient (Figure 3). In only one of ELS patients, partial absence of the pericardium was observed as a congenital abnormality (Figure 4). Blood supply to the pulmonary sequestration originated from the thoracic aorta in 19 patients (59.4%), the abdominal aorta in nine patients (28.1%), the intercostal artery in two patients (6.3%), the thoracic aorta and phrenic artery in one patient (3.1%), and the bronchial and phrenic arteries in one patient (3.1%). Venous drainage was through the inferior pulmonary vein in 27 patients (84.4%), the superior pulmonary vein in three patients (9.4%), the left atrium in one patient (3.1%), and the brachiocephalic vein in one patient (3.1%). Lobectomy was performed in 17 (53.1%), wedge resection in eight (25%), mass excision in four (12.5%), and segmentectomy in three (9.4%) patients for pulmonary sequestration (Table 2). Postoperative histopathological examinations confirmed the diagnosis of pulmonary sequestration and excluded any other diagnosis in all patients.

Table 1. Symptoms, diagnostic methods and surgical procedures

	n	%
Symptoms		
Cough	21	65.6
Voluminous sputum expectoration	8	25
Hemoptysis	6	18.8
Dyspnea	6	18.8
Chest pain	3	9.4
Asymptomatic	7	21.9
Diagnostic method		
Chest X-ray (only)	9	
Computed tomography (only)	12	
Computed tomographic angiography	4	
Digital subtraction angiography	5	
Magnetic resonance angiography	2	
Surgical procedures		
Lobectomy	17	53.1
Segmentectomy	3	9.4
Wedge resection	8	25
Mass excision	4	12.5
Total	32	100

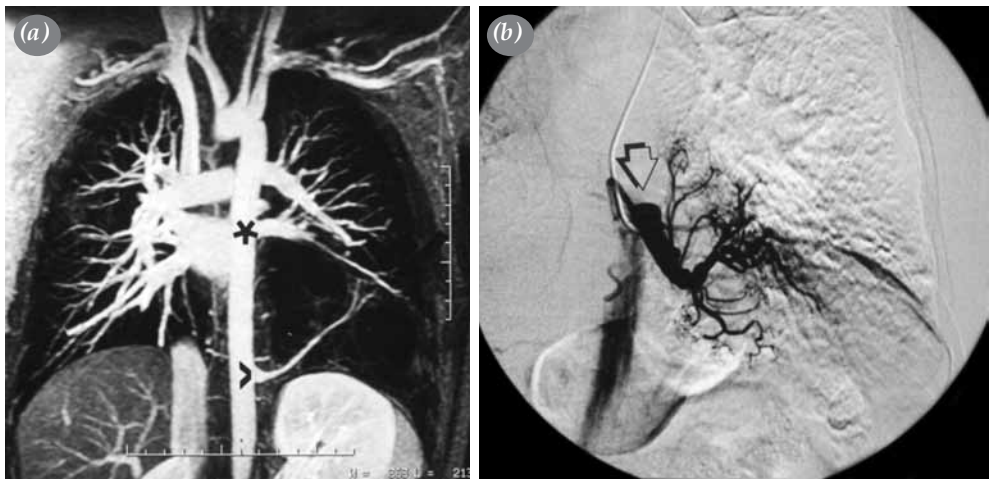

Figure 1. (a) Maximum intensity projection view of 3-dimensional contrast enhanced magnetic resonance angiography; aberrant artery arising from thoracic aorta (arrow head), which was supplying pulmonary sequestration and venous drainage into inferior pulmonary vein (star). **(b)** Digital subtraction angiography demonstrated a large aberrant artery, measuring 2 cm diameter at its origin, arising from distal thoracic aorta (arrow head).

Postoperative complications occurred in six patients (18.8%) as prolonged air leak (longer than 10 days) in three patients, wound infection in two patients, and empyema in one patient. The wound infections were easily managed with the appropriate antibiotics. The empyema was treated with tube drainage and effective antibiotic was administered. No mortality was observed. Median follow-up time, which was completed in 24 patients (75%), was 42 months (range 3 to 105 months) and all patients remained asymptomatic.

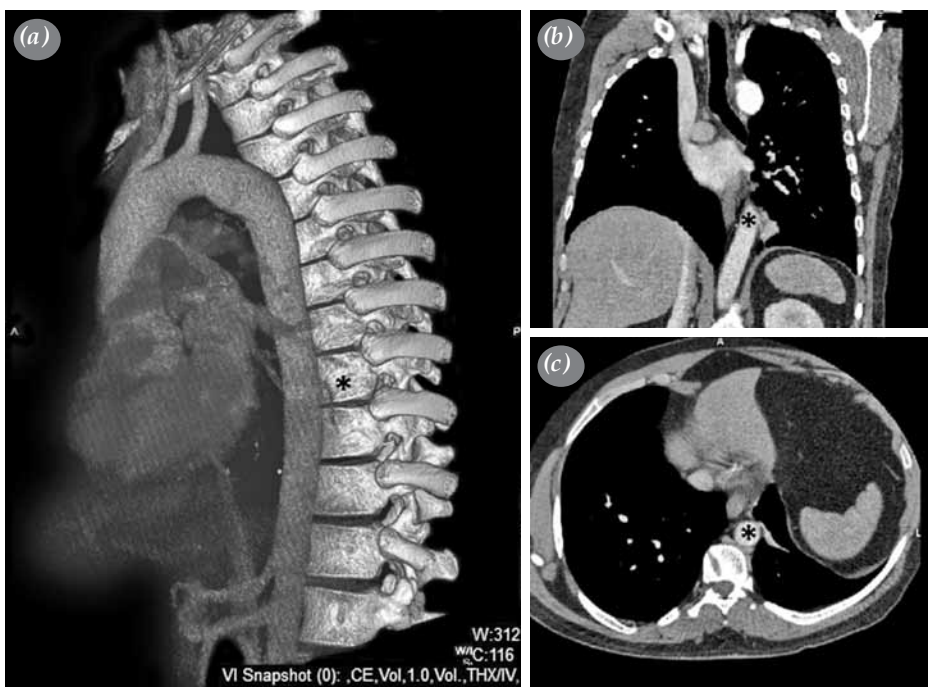

Figure 2. Aberrant artery arising from thoracic aorta (star), which was supplying pulmonary sequestration: **(a)** Maximum intensity projection view of 3-dimensional contrast enhanced computed tomographic angiography; **(b)** coronal view; **(c)** axial view.

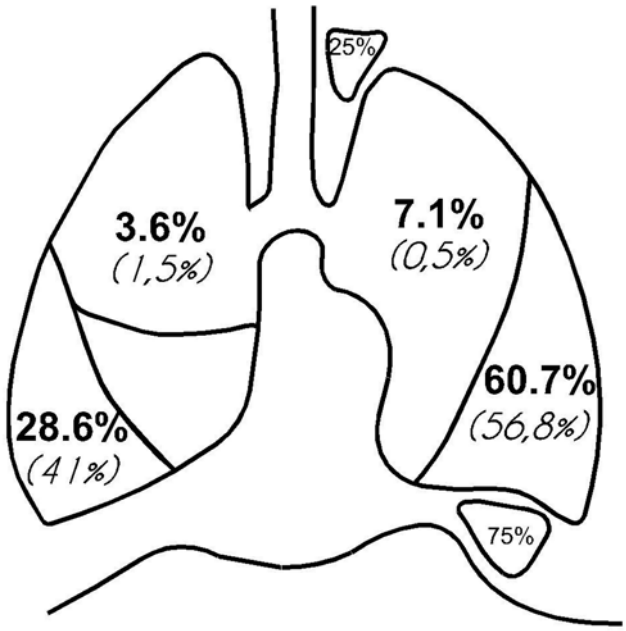

Figure 3. Distribution of intralobar sequestration or extralobar sequestration cases in our series. Numbers in parentheses belong to Savic's series of 540 cases.[1]

DISCUSSION

Pulmonary sequestration is a rare bronchopulmonary foregut malformation. The term 'sequestration' was introduced by Pryce in 1946.[3] Numerous theories have been suggested regarding its pathogenesis, but all remain controversial. A sequestration probably arises from lung buds as an independent part with its own blood supply during the separation of the trachea and lung buds from the foregut.[4]

Cough, voluminous sputum expectoration, recurrent episodes of pneumonia, hemoptysis and even congestive heart failure are the clinical presentations of pulmonary sequestration. Since these symptoms are not typical, pulmonary sequestration may remain disguised for years and is often misdiagnosed as bronchiectasis, pneumonia and even mass lesion. Patients may also be asymptomatic and pulmonary sequestration may be diagnosed incidentally.[5,6] In this series, 25 patients (78.1%) were symptomatic whilst 21.9% (all ELS patients and three ILS patients) was asymptomatic.

Extralobar sequestration is commonly associated with congenital malformations including congenital heart disease, congenital diaphragmatic hernia, chest wall, vertebra and other congenital pulmonary abnormalities, such as hypoplasia, congenital cystic adenomatoid malformation, congenital lobar emphysema or bronchogenic cyst.[1,2,7] The most common coexisting abnormality is congenital diaphragmatic hernia.[8] In this series, one of the ELS was associated with partial absence of the pericardium. In contrast to extralobar sequestration, intralobar sequestration is infrequently associated with other congenital abnormalities.[9]

Halkic et al.[10] stated that ILS often remains unrecognized until after the age of 20. The current study data support this statement with the mean age of 33.3±13.7 years (range 4 to 61 years) at diagnosis for ILS patients. Intralobar sequestration rarely causes problems in childhood.[1,11] The diagnosis was established at the age of four years in only two of the ILS patients in the current study. Intralobar

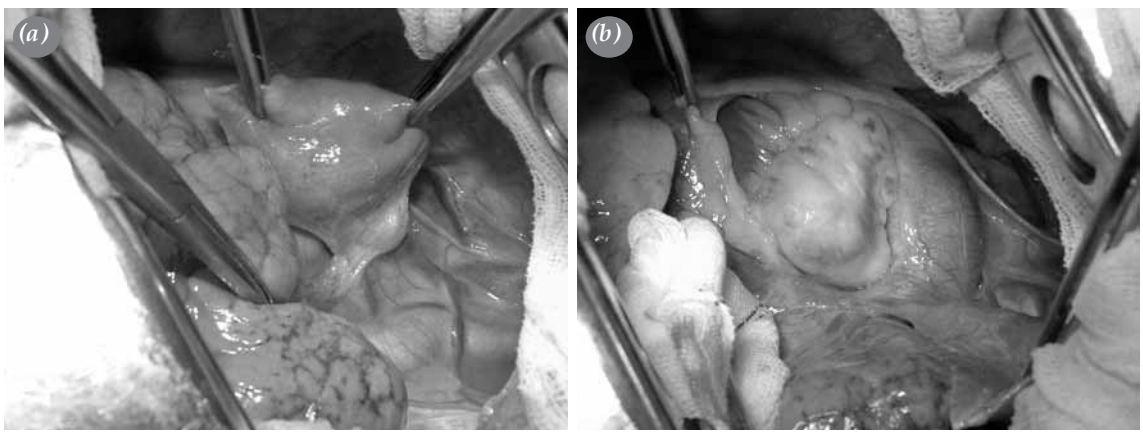

Figure 4. (a) At intraoperative view; extralobar sequestration which was located at left side, receiving its blood supply from arcus aorta and venous drainage through brachiocephalic vein. **(b)** Congenital pericardial defect of 5x5 cm in dimension.

Table 2. Characteristics of patients, sequestrations and surgical procedures

Case	Age/Gender	Type	Side	Location	Blood supply	Venous drainage	Resection
1	30/F	ILS	R	RL	Abdominal aorta	Inferior pulmonary vein	Lobectomy
2	25/F	ILS	L	LL	Intercostal artery	Inferior pulmonary vein	Wedge resection
3	23/F	ILS	L	LL	Abdominal aorta	Inferior pulmonary vein	Wedge resection
4	21/F	ILS	L	LL	Intercostal artery	Inferior pulmonary vein	Wedge resection
5	25/M	ILS	R	RU	Thoracic aorta	Superior pulmonary vein	Segmentectomy
6	19/M	ELS	L	Base	Thoracic aorta	Inferior pulmonary vein	Excision
7	43/F	ELS	L	Base	Abdominal aorta	Inferior pulmonary vein	Excision
8	32/M	ELS	L	Base	Thoracic aorta	Left atrium	Excision
9	29/M	ILS	L	LU	Thoracic aorta	Superior pulmonary vein	Wedge resection
10	43/M	ILS	L	LU	Thoracic aorta	Superior pulmonary vein	Wedge resection
11	26/M	ILS	R	RL	Abdominal aorta	Inferior pulmonary vein	Segmentectomy
12	21/M	ILS	L	LL	Abdominal aorta	Inferior pulmonary vein	Lobectomy
13	48/F	ILS	R	RL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
14	37/F	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
15	33/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
16	4/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
17	30/F	ILS	R	RL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
18	32/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
19	4/F	ILS	R	RL	Abdominal aorta	Inferior pulmonary vein	Lobectomy
20	51/M	ILS	L	LL	Thoracic aorta + phrenic artery	Inferior pulmonary vein	Lobectomy
21	58/M	ILS	L	LL	Bronchial + phrenic artery	Inferior pulmonary vein	Lobectomy
22	32/F	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
23	32/F	ILS	L	LL	Abdominal aorta	Inferior pulmonary vein	Lobectomy
24	19/F	ELS	L	Apex	Thoracic aorta	Brachiocephalic vein	Excision
25	43/F	ILS	R	RL	Abdominal aorta	Inferior pulmonary vein	Wedge resection
26	24/M	ILS	L	LL	Abdominal aorta	Inferior pulmonary vein	Lobectomy
27	61/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Wedge resection
28	26/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
29	41/F	ILS	R	RL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
30	31/F	ILS	R	RL	Thoracic aorta	Inferior pulmonary vein	Lobectomy
31	32/M	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Segmentectomy
32	50/F	ILS	L	LL	Thoracic aorta	Inferior pulmonary vein	Wedge resection

F: Female; M: Male; ILS: Intralobar sequestration; ELS: Extralobar sequestration; R: Right; L: Left; RL: Right lower; LL: Left lower; RU: Right upper; LU: Left upper.

sequestration has almost equal gender prevalence.[9] In the current study, distribution between the genders was equal as reported in literature.

Intralobar sequestration is contained within the normal pulmonary parenchyma without its own visceral pleura.[5] Intralobar sequestration constitutes three-quarters of all pulmonary sequestrations. Typically, ILS is located in the lower lobes, and the posterior basal segment is most often involved.[1,5,10] In the current series, 87.5% of cases were ILS. In 25 patients (89.3%), the ILS was located in the lower lobes, and in the remaining three patients (10.7%) in the upper lobes. This rate is higher than the 2% reported by Savic et al.[1] for upper lobes.

Extralobar sequestration is usually diagnosed in infancy or childhood.[10] Extralobar sequestration may be present shortly after birth with respiratory distress or feeding difficulties. It may be diagnosed at the time of diaphragmatic hernia repair or during screening for another congenital abnormality, incidentally.[7] It has also been reported that ELS may remain asymptomatic for many years.[3] The mean age of the patients with ELS in the current study was 28.3±11.6 years (range 19 to 43 years). Even though a few reports have suggested no sex predominance for ELS, many authors have reported that ELS occurs three to four times more often in males than in females.[1,7] However, no gender predominance was recorded in the current study.

Extralobar sequestration is separated from normal pulmonary parenchyma with its own visceral pleura. Nearly one-quarter of sequestrations are ELS.[2,12] Extralobar sequestration is frequently located between the left lower lobe and the diaphragm.[5,8] In the current series, there were only four patients (12.5%) of ELS, of which three were located at the base of the hemithorax and one was at the apex.

Blood supply to the pulmonary sequestration is almost always from a systemic artery. Savic et al.[1] stated in their study of 540 patients that the blood supply of both ILS and ELS was from the thoracic aorta in 68.5%, the abdominal aorta in 21.3%, and the intercostal artery in 3.4% of cases. Halkic et al.[10] reported blood supply from the thoracic aorta in 80% and the abdominal aorta in 20% of cases. In the current series, the main blood supply to the sequestrated part arose from the thoracic aorta in 65.6%, the abdominal aorta in 28.1%, and the intercostal artery in 6.3% of patients as in the series above. The distribution for the ELS cases in detail was three from the thoracic aorta and one from the abdominal aorta.

Venous drainage is always through pulmonary veins in ILS and through a systemic vein in ELS.[2,5,13] In all the ILS patients of the current study, venous drainage was through pulmonary veins (three superior and 25 inferior pulmonary veins). Of the ELS patients, venous drainage was through the pulmonary veins in two patients, the left atrium in one patient, and the brachiocephalic vein in one patient.

A wide variety of imaging techniques have been used in the diagnosis of pulmonary sequestration, including routine chest radiography, conventional tomography, radionuclide scanning, bronchography, and more recently ultrasonography, CT and magnetic resonance imaging. Traditionally, the diagnosis of pulmonary sequestration has been made definitively with arterial angiography.[14] Digital subtraction angiography, CTA, and MRA are the most useful diagnostic methods, which can display the arterial and venous vascular anatomy of pulmonary sequestration.[6] Recently, CTA is an effective non-invasive method for confirming the correct diagnosis of pulmonary sequestration with high accuracy, sensitivity and specificity of 97.7%, 97.3% and 100%, respectively. It is faster and can display not only the vascular anatomy but also provide detailed information about the bronchial anatomy and the pulmonary parenchyma.[15] Chest radiography was the primary diagnostic method for all the patients in this series, and the only imaging technique in nine patients owing to the absence of CT before 1993 in our institution. In the remaining 23 patients (71.9%), thoracic CT was applied. For further confirmation, five patients underwent DSA, four underwent CTA, and two underwent MRA. Pulmonary sequestration was diagnosed with preoperative diagnostic methods in 15 patients (46.9%) and the diagnosis could be verified after histopathological examination of the resected specimen in the remaining 17 patients (53.1%). Since we have begun to consider that CTA should be carried out for cases that are difficult to diagnose, we expect that the rate of preoperative definitive diagnosis will increase with the decisive role of CTA in the diagnosis of pulmonary sequestration.

The definitive treatment of pulmonary sequestration is surgical resection. Recently, therapeutic embolization has been reported as an alternative method to surgical resection especially in neonates and children.[16,17] Goto et al.[18] reported coil embolization as a safe, feasible, and effective method before surgery to prevent intraoperative bleeding from aberrant arteries. Neither therapeutic nor preventive coil embolization was performed in our clinical practice.

Resection of sequestrated parenchyma can be performed via thoracotomy or video assisted thoracoscopic surgery (VATS). Although Wan et al.[19] first described VATS lobectomy for the surgical treatment of pulmonary sequestration in 2002, the advantages of VATS for pulmonary sequestration resection are still controversial and need further research. In a study comparing VATS and posterolateral thoracotomy for pulmonary sequestration resection, Liu et al.[13] reported no significant differences between the two groups in terms of the duration of operation, blood loss, amount of chest drainage, duration of chest drainage, length of postoperative hospital stay, and occurrence of complications. Conversely, Sun and Xiao[20] reported that patients undergoing VATS had a statistically significantly shorter length of postoperative hospital stay than patients undergoing thoracotomy, indicating that VATS might be preferable to thoracotomy for pulmonary sequestration resection. Although embolization as an interventional therapy and VATS seem more patient-friendly and less invasive, we have not yet had appropriate patients to present any findings.

Removal of sequestrated areas of the pulmonary parenchyma is the mainstay of the surgical management. Since preservation of the functioning pulmonary parenchyma is an important consideration, we performed not only segmentectomy, but also wedge resection in cases where the entire lobe was not involved. In fact, wedge resection was preferred rather than segmentectomy for this purpose (eight wedge resections and three segmentectomies).

The pathological findings include dilated bronchioles, alveolar ducts and alveoli, interstitial fibrosis and thickened vasculature with chronic inflammation surrounding dilated airspaces lined by respiratory epithelium. Concomitant acute and organizing pneumonia may be present on histological

appearance.[21] Postoperative histopathological examinations confirmed the diagnosis of pulmonary sequestration and excluded any other diagnosis in all the current cases.

Even though pulmonary sequestration is benign in nature, its clinical course may be severe and life-threatening depending on complications of congestive heart failure, hemoptysis, pneumonia, hemothorax, and malignant degeneration.[3,22,23] Hence, we support the decision of surgical removal of pulmonary sequestration even for asymptomatic patients.

In conclusion, it is crucial to consider pulmonary sequestration in the differential diagnosis of recurrent infections and hemoptysis. Currently, it may be easy to diagnose pulmonary sequestration with advanced radiological methods. Computed tomographic angiography is a surgeon-friendly method in the management of pulmonary sequestrations to evaluate the pulmonary parenchyma, display vascular and bronchial anatomy, and rule out other pathologies.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

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