

Clinical Features and Management of Bronchogenic Cysts: Report of 17 Cases

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Abstract: Between 1966 and 1996, 17 patients, comprising 1 child and 16 adults, underwent surgical treatment for bronchogenic cysts at the National Cancer Center Hospital. The bronchogenic cysts were located in the mediastinum in 11 patients and in the pulmonary parenchyma in 6. Of the 17 patients, 5 (29.4%) manifested symptoms, being more frequently seen in those with intrapulmonary cysts than in those with mediastinal cysts. Chest radiographs were ineffective for accurate preoperative diagnosis, but accurate diagnosis was possible with 69.2% of computed tomography (CT) scans and 100% of magnetic resonance imaging (MRI) scans. MRI also proved very useful for qualitatively diagnosing the mediastinal tumors as cystic or solid. Surgery was performed through a thoracotomy in 14 patients and by video-assisted thoracic surgery (VATS) in 3 patients, achieving complete resection in 16 patients. In one patient, a mediastinal bronchogenic cyst was excised by VATS and incompletely resected because of tight adhesion to the membranous part of the trachea; however, no late complication or recurrence developed after the residual cystic wall had been ablated by electrocautery. VATS, which is an easy procedure to perform with only minimal surgical invasion, may be indicated for bronchogenic cysts if patients who undergo incomplete resection can be followed up carefully. Recent advances in imaging techniques have made it unnecessary to perform surgical excision for diagnostic confirmation, but we recommend surgery for most patients to relieve symptoms and prevent complications.

Key Words: bronchogenic cyst, mediastinal cyst, computed tomography, magnetic resonance imaging, video-assisted thoracic surgery

Introduction

Bronchogenic cysts are thought to originate from abnormal budding of the tracheal diverticulum between the third and sixth weeks of gestation. They develop as part of the tracheobronchial tree, separate from the primary airway as a result of aberrational development,¹ and are usually found in the mediastinum around the tracheobronchial tree or in the pulmonary parenchyma. Recent studies suggest that these cysts frequently cause symptoms in adults, and therefore surgical excision is recommended to prevent complications and avoid the operative difficulties associated with resecting symptomatic cysts.² However, after the diagnosis of a bronchogenic cyst has been established, making a decision about whether to remove the cyst surgically may be difficult, especially in an asymptomatic patient.

We present herein a retrospective review of 17 patients with bronchogenic cysts treated at the National Cancer Center Hospital over the last 30 years, and discuss the indications for their treatment.

Patients and Methods

The operative records and pathological reports of 22 patients who underwent resection of a bronchogenic cyst at our institution during the 30-year period from 1966 to 1996 were reviewed. After the exclusion of 5 patients on morphologic grounds because their cysts were not clearly distinguished from an abscess or other chronic cavity, such as an infected sequestration, the remaining 1 child and 16 adults formed the study population. These patients were divided into two groups according to whether they had mediastinal cysts or intrapulmonary cysts, to facilitate analysis of the results according to the cyst location. Data on demographics, symptoms, imaging, surgical procedures, and postoperative complications were collected for each group.

Table 1. Clinical characteristics of the 17 patients with bronchogenic cysts

	Mediastinal (n = 11)	Intrapulmonary (n = 6)	Significance
Average age (years)	34.6 ± 16.9	54.7 ± 15.7	<i>P</i> < 0.05
Sex (M/F)	5/6	4/2	NS
Average size (cm)	4.2 ± 1.4	4.3 ± 1.7	NS
Symptomatic	1 (9.1%)	4 (66.7%)	<i>P</i> < 0.01
Mode of presentation			
cough	—	4	
sputum	—	3	
pain	1	1	
fever	—	2	
weight loss	—	1	

NS, not significant

Follow-up data were obtained from the office records. Statistical comparisons were carried out using the unpaired Student's *t*-test or Fisher's exact test, and differences of *P* < 0.05 were considered to be significant.

Results

There were 9 males and 8 females, ranging in age from 4 to 69 years with an average age of 40.8 ± 18.5 years.

Location and Clinical Manifestation

The characteristics of the patients according to the location of the lesion are summarized in Table 1. There were 11 patients with mediastinal cysts and 6 with intrapulmonary cysts. The locations of the mediastinal cysts were classified according to Maier,³ five being paraesophageal, three carinal and paratracheal, and one hilar. There were three bronchogenic cysts located in the anterolateral aspect of the thoracic vertebral bodies. Of the six intrapulmonary cysts, three were located in the left upper lobe, one in the right upper lobe, and two in the right lower lobe. The intrapulmonary cysts had no particular locational predilection.

Of the 17 patients, 5 (29.4%) had presented with symptoms, the most common of which was coughing, seen in 4 patients, 3 of whom had purulent sputum associated with pneumonia. The other symptoms included pain in 2 patients, fever in 2, and weight loss in 1. Most of the symptomatic patients had two or more symptoms. With regard to the incidence of symptoms according to cyst location, 4 (66.7%) of the 6 patients with intrapulmonary bronchogenic cyst had symptoms, compared with only 1 of the 11 (9.1%) patients with mediastinal cysts. The incidence of symptoms differed significantly (*P* < 0.01) between the patients with mediastinal cysts and those with intrapulmonary cysts.

Table 2. Results of imaging studies

	Chest X-ray	CT scan	MRI
No. of Cases	17	13	4
Abnormal findings	16	13	4
Preoperative diagnosis			
cyst	0	9	4
benign Tumor	13	4	—
malignant Tumor	1	—	—
other	2	—	—

CT, computed tomography; MRI, magnetic resonance imaging

Imaging

Table 2 shows the sensitivity and clinical use of the imaging techniques of X-ray, computed tomography (CT), and magnetic resonance imaging (MRI). Abnormal shadows were detected by chest X-ray in 16 (94.1%) of the 17 patients, but this test was of little diagnostic value. One of the shadows was mistaken for a malignant tumor because of its ill-defined contours.

CT was performed in 13 patients and proved useful for delineating the cyst location in relation to the adjacent structures; however, CT was of limited value for analyzing the internal properties of the cysts. Only 9 patients (69.2%) were shown to have homogeneous isodensity with water, suggestive of a benign cyst, whereas high density suggestive of a solid tumor was evident in 4 patients (30.8%).

Four patients in this series were examined by both CT and MRI, and bronchogenic cysts were diagnosed preoperatively in all of them by both imaging techniques. A comparison of these two diagnostic techniques is shown in Table 3. We measured the CT density of the bronchogenic cysts in 2 of these 4 patients. Higher CT numbers were found in both of these lesions, but we were able to diagnose them as bronchogenic cysts by MRI

Table 3. Comparison of CT and MRI findings

No.	Location	CT		MRI		
		CT Number	Diagnosis	T1-Weighted	T2-Weighted	Enhancement
1	Posterior mediastinum	60	Benign tumor	Iso	High	—
2	Posterior mediastinum	31	Benign tumor	Low	High	—
3	Subcarina	Not calculated	Cyst	High	High	—
4	Subcarina	Not calculated	Cyst	Iso-high	High	—

imaging, which showed high signal intensity on T2-weighted images and lack of enhancement following the injection of gadolinium contrast medium. Two cysts showed iso-intensity on T1-weighted images and high signal intensity on T2-weighted images, and another cyst showed high signal intensity on both T1- and T2-weighted images. Overall, 3 of the 4 patients had iso- or high signal intensity on T1-weighted images, and only one showed low signal intensity, indicating a pure serous cyst, on T1-weighted images.

Surgical Procedures, Operative Findings, and Complications

Surgical resection through a standard thoracotomy was performed in 6 patients with intrapulmonary cysts and 8 with mediastinal cysts. Towards the end of the series, resection by video-assisted thoracic surgery (VATS) was carried out for three mediastinal bronchogenic cysts, but pulmonary resection was required for all the intrapulmonary cysts. Lobectomy and segmentectomy were performed in 5 patients and 1 patient, respectively. All the intrapulmonary cysts were completely resected.

In five of the six intrapulmonary cysts, marked pleural adhesions were found, and two of the mediastinal cysts showed adhesion, but not communication, to the esophageal wall or membranous portion of the trachea. These adhesions created problems during surgery. One mediastinal cyst closely adherent to the esophageal wall was excised by dissociating the muscular fibers of the esophagus, and the defect in the muscular fibers was simply sutured without any complications (Table 4).

Thoracoscopic excision of three mediastinal bronchogenic cysts was successfully performed; however, the excision was incomplete in one patient (no. 2) whose cyst, located in the subcarinal area, was covered by a wall partly composed of the membranous portion of the trachea. The residual wall was ablated using electrocautery (Table 5).

Pathological Findings

The cysts varied in size from 2 to 9 cm in diameter, with an average diameter of 4.2 ± 1.4 cm. Of the 17 bron-

Table 4. Operative procedures, findings, and complications

	Mediastinal (<i>n</i> = 11)	Intrapulmonary (<i>n</i> = 6)
Procedures		
thoracotomy	8	6
VATS	3	0
Operative Findings		
adhesion to a neighboring organ	2	0
pleural adhesion	0	5
Complications		
intraoperative	1	0
postoperative	0	0

VATS, video-assisted thoracic surgery

chogenic cysts, 16 were unilocular and one mediastinal cyst was multilocular. None of the mediastinal or intrapulmonary cysts communicated with the tracheobronchial tree. The contents of the cysts showed a great variety of color and consistency, and inflammatory changes were found in three of the intrapulmonary cysts. All of the cysts had a ciliated columnar epithelial lining as a typical histological feature. Epithelial hyperplasia and squamous metaplasia were observed in one intrapulmonary cyst and one mediastinal cyst. Islets of cartilage were present in seven mediastinal cysts and in three intrapulmonary cysts (Table 6).

Follow-Up

The follow-up period ranged from 2 months to 13 years, with an average period of 30.3 ± 41.4 months. No symptoms or evidence of recurrence were found in any of the patients after resection.

Discussion

Mediastinal bronchogenic cysts account for 5%–10% of all mediastinal tumors, and are differentiated as mediastinal bronchogenic cysts or intrapulmonary bronchogenic cysts according to the embryonic phase that marks the development of an abnormal bronchial primordium.⁴ While various reports indicate that media-

Table 5. Summary of patients who underwent resection of a bronchogenic cyst by VATS

Patient no.	Sex	Age (years)	Symptoms	Preoperative diagnosis	Operation time	Postoperative hospital stay (days)	Resection	Complications
1	M	27	None	Benign tumor	1 h 02 min	8	Complete	None
2	M	4	None	Cyst	1 h 55 min	2	Incomplete	None
3	F	40	None	Cyst	1 h 45 min	4	Complete	None

Table 6. Pathological findings

	Mediastinal (<i>n</i> = 11)	Intrapulmonary (<i>n</i> = 6)
Unilocular	10	6
Multilocular	1	0
Communication with tracheobronchial tree	0	0
Evidence of infection	0	3
Epithelial hyperplasia and/ or squamous metaplasia	1	1
Islets of cartilage	7	3

stinal cysts or intrapulmonary cysts predominate,⁵⁻⁷ Rogers and Osmer⁶ and Ramenofsky et al.⁷ claim that the incidence of intrapulmonary cysts is higher. Conversely, in our series, the incidence of mediastinal cysts was higher.

Bronchogenic cysts are benign tumors that cause a variety of symptoms and complications. Recent reports have stated that a large number of bronchogenic cysts in adults are symptomatic and that a high proportion of asymptomatic cysts also become symptomatic during the course of observation.^{2,5,7} Among the patients treated at our facility, 29.4% were symptomatic, a lower proportion than that reported by many researchers, but by no means a small number.

Preoperative diagnosis is established primarily by chest X-ray and CT, but MRI has also come into recent use. Although plain X-ray examination can detect the localized presence of a tumor and a differential diagnosis can be inferred from frequent foci of occurrence, this technique alone cannot visualize the characteristic features of bronchogenic cysts. In comparison, CT is deemed useful for ascertaining the localized presence and properties of a tumor, but the rate of correct preoperative diagnosis is relatively low at 45%–61%, even with the use of CT.^{2,5} In our series, only 69.2% of the bronchogenic cysts were accurately diagnosed due to the fact that CT is inadequate for diagnostic imaging of features within a bronchogenic cyst. As for the nature of the mass, density and the heterogeneous aspect can be sources of error, as most bronchogenic cysts have a low CT number of 0 to 20 Hounsfield units, although occasionally, the cysts contain turbid mucoid fluid resulting in high CT numbers and may appear solid.⁸

MRI is an extremely useful technique for providing supplementary diagnostic data. In a typical noninfected cyst containing serous fluid, T1-weighted images show a low signal, and T2-weighted images show a high signal; however, in reality, bronchogenic cysts rarely present such a pattern. Murayama et al.⁹ reported high T1-weighted image signals in five out of five bronchogenic cysts, and these results are thought to reflect the intracystic protein component, mucosal component, or hemorrhage.¹⁰ In our study, two lesions considered to be solid tumors according to the CT findings were subsequently diagnosed as bronchogenic cysts through the use of MRI. The T1-weighted images showed a moderate to high signal in three of the four lesions examined by MRI in this series, all of which were diagnosed as cysts because they showed both a high T2-weighted signal and no contrast effect upon contrast-enhanced MRI. Thus, even when CT suggests a solid tumor, we believe that an almost precise essential diagnosis can be made preoperatively, without reference to T1-weighted image signal strength, when the T2-weighted signal is high and contrast-enhanced MRI shows no contrast.

In recent years, surgical excision has been advocated for bronchogenic cysts.^{2,5} There is general agreement concerning surgical treatment for symptomatic patients; however, the most appropriate treatment for asymptomatic patients remains controversial. The factors for consideration when contemplating surgery for asymptomatic patients include the limits of diagnostic imaging, the possibility of symptomatic manifestations with time,^{2,5} and the possibility of malignant transformation of the cyst.¹¹ The frequency of symptomatic manifestation during the observation period varies from 23% to 72%.^{2,12} It is difficult to appreciate the exact prevalence of symptomatic cysts, one reason for which is that a bias should be applied to patient selection factors, and also the advanced age of some patients proves that these cysts can remain silent forever. Furthermore, we must bear in mind that surgery is by no means easy because of complicating anatomic factors once symptoms arise. Thus, in consideration of their high incidence of complications,² lesions presumed to be bronchogenic cysts should usually be removed. With regard to malignant transformation, a causal relationship between cysts and malignant tumors remains unclear in most reported

casts,^{13,14} although Okada et al.¹¹ and Miralles et al.¹⁵ both presented convincing evidence that a malignancy had arisen from a congenital bronchogenic cyst in two adults.

Occasional reports of increasingly noninvasive thoracoscopic resection for mediastinal cysts have recently been described, these procedures being considered to allow early discharge.¹⁶ Other reports state that even following incomplete excision, recurrence can be prevented by ablating the residual tissue with electrocautery.¹⁷ In our series, we performed three thoracoscopic resections of mediastinal bronchogenic cysts; however, in one patient, strong adhesion to the trachea necessitated incomplete excision that left the adhering cystic wall behind, the mucosal surface of which was thoroughly cauterized with an electric knife. No signs of recurrence have been observed in this patient during 3 years of postoperative follow-up. There is some opinion that complete excision is nevertheless essential to prevent recurrence,^{5,18} and that the introduction of thoracoscopic surgery will require even longer-term follow-up observation to prove the validity of the technique.

There is no dispute about the need for surgical intervention for symptomatic bronchogenic cysts; however, the indications of surgery for asymptomatic cysts are controversial. Based on the large number of patients that present with symptoms, the difficulty in performing surgery once inflammation has developed, and the occasional but significant reports of malignant transformation, our position at present is to plan elective excision of lesions diagnosed by imaging techniques as bronchogenic cysts.

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