Mediastinal Bronchogenic Cysts: Clinical Presentation, Diagnosis, and Treatment Outcomes

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Abstract

Background and aims: Bronchogenic cysts are rare mediastinal tumors caused by foregut malformations. Although surgery remains the definitive form of diagnosis and treatment, we can reach a diagnosis by imaging modalities. This retrospective study aims to analyze our experience with bronchogenic cysts and present a discussion of the demographics of patients, patient signs and symptoms, and cyst complications, as well as a surgical approach for resection and surgical outcomes for patients described in this study.

Methods: This retrospective, descriptive cross-sectional study reviewed the medical records of 12 patients who were formally diagnosed with bronchogenic cysts by histopathology and treated surgically between 2010 and 2020. We reviewed the medical records of all patients, including age, location of the cyst, symptoms, complications, imaging techniques, and surgical interventions.

Results: In total, twelve bronchogenic cyst cases were included: eight mediastinal cysts (two intrathymic) and four intraparenchymal cysts. One patient was asymptomatic, and the remaining 11 were symptomatic. The most common symptoms were chest pain, dyspnea, and cough. Four cases suffered from severe bronchogenic cyst complications, of which three had pneumonia and one had atelectasis. The longest axis of a bronchogenic cyst ranged from 2–11cm (mean = 4.52cm). All 12 patients underwent complete surgical resection of the cyst without postoperative complications or recurrence.

Conclusion: Although bronchogenic cysts are rare, they should be considered in the differential diagnosis of mediastinal tumors. In both symptomatic and asymptomatic cases, complete surgical resection is the best option to prevent future recurrence and complications, such as malignancy.

Keywords: Mediastinum, bronchogenic cyst, thoracotomy

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INTRODUCTION

Although bronchogenic cysts are rare, they remain the most common type of mediastinal cyst, which accounts for 12–18% of all mediastinal tumors [1]. Therefore, the correct diagnosis and management of bronchogenic cysts is extremely important. A bronchogenic cyst is a respiratory tract malformation that originates from the foregut [2]. They can be found anywhere along their pathway of development in an ectopic site, but most are in the

mediastinum, mainly in the middle and posterior mediastinum in the early gestational stage or in the later stage in the lung [2, 3].

Although with the development of imaging modalities, bronchogenic cysts have been diagnosed more frequently in recent years [4], a definitive diagnosis can only be made through histopathological studies after complete excision of the cyst through surgery [3]. The preoperative diagnosis of such cysts is mostly performed using CT or MRI, but they are not always accurate because they can simulate multiple lesions and are difficult to diagnose by biopsy [4, 5]

Most cases are asymptomatic and diagnosed incidentally by imaging, although they may become abnormally large and cause coughing, chest pain,

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dyspnea, and dysphagia [6]. Cysts may also become infected with gram-positive and gram-negative bacteria, resulting in purulent expectoration/sputum and fever [7]. Therefore, surgical treatment of bronchogenic cysts is strongly recommended, due to its association with lower mortality and morbidity when compared with conservative treatment and observation, and it has an extremely low recurrence rate [1].

This retrospective study aims to review and analyze the cases of 12 patients formally diagnosed with a bronchogenic cyst at Jordan University Hospital from 2010 to 2020, two of which are intrathymic cysts, which are rare [3]. We review the age, gender, symptoms, and locations of the cysts and investigate the complications and recurrence rate of bronchogenic cysts to show the presentations of patients with bronchogenic cysts more comprehensively and accurately.

SUBJECTS AND METHODS

This is a retrospective, descriptive, and cross-sectional study. The medical records of 12 patients with bronchogenic cysts surgically resected at Jordan University Hospital between 2010 and 2020 were reviewed. Histopathologic diagnosis of the bronchogenic cyst was used as the inclusion criterion.

The patient's age, gender, symptoms, tumor size and location, type of surgery used, postoperative complications, follow-up data, imaging modalities and descriptions, and histopathological results were all obtained from the patient's medical records after obtaining approval from the ethical committee (Table 1). Patient confidentiality was maintained.

Microsoft Excel was used to analyze data and measure the mean, median, mode, and percentage to describe and compare different values and results.

Table 1: Patient demographics, symptoms, cyst location, complications and surgical approach

	Age in vears/sex	Location	Symptoms	Complications of bronchogenic cyst	Surgical treatment
1	0.25 /F	Posterior mediastinum	Dyspnea	bronchogeme cyst	Thoracotomy
1			V 1		,
2	43 /F	Anterior mediastinum (intrathymic)	Chest pain, cough		Sternotomy
3	37 /M	Posterior mediastinum	Chest pain, cough, dysphagia	pneumonia	Thoracotomy
4	40 /M	posterior mediastinum	Chest pain, cough, dyspnea	atelectasis	Thoracotomy
5	53 /F	Anterior mediastinum	Chest pain, dyspnea		Thoracotomy
6	58 /F	Right lower lobe	Cough, hemoptysis		Thoracotomy
7	58 /M	Anterior mediastinum (intrathymic)	Asymptomatic		Sternotomy
8	48 /F	Right lower lobe	Chest pain, dyspnea		Thoracotomy
9	54 /F	Anterior mediastinum	Chest pain		Thoracotomy
10	13 /M	Left lower lobe	Cough	pneumonia	Thoracotomy
11	29 /F	Posterior mediastinum	Cough, dyspnea		Thoracotomy
12	1.58/ M	Right upper lobe	Cough, dyspnea	pneumonia	Thoracotomy

RESULTS

Findings and location

Over ten years (2010–2020), 12 patients (five males and seven females, with a male to female ratio of 1:1.4), aged 3 months to 58 years (mean age = 36.23 years) underwent surgical excision of bronchogenic cysts of the mediastinum and lungs at Jordan University Hospital. Of these mediastinal bronchogenic cysts (n=8), four were in the anterior mediastinum (two intrathymic) and four were in the posterior mediastinum. There were four intraparenchymal bronchogenic cysts with two in

the right lower lobe, one in the right upper lobe, and one in the left lower lobe (Table 1).

Clinical presentation

One patient (8.3%), aged 58, was asymptomatic at the time of the discovery of the bronchogenic cyst, as it was found incidentally during an aortic valve replacement surgery. Eleven patients (91.7%) were symptomatic at presentation with a mean age of 34.25 years. Most patients had more than one symptom. The most common symptoms were chest pain, dyspnea, and coughing (Table 2).

Table 2: P	resenting sy	mptoms of	the	patients
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Symptom	Number	Percent %
Chest pain	6	50%
Dyspnea	6	50%
Cough	6	50%
Dysphagia	1	8.3%
Hemoptysis	1	8.3%

Four patients (33.3%) suffered from severe complications due to bronchogenic cysts (mean age 22.89 years). All four were male patients, among whom were three cases of pneumonia and one of atelectasis (Table 1).

For diagnosis, various imaging modalities were performed on all patients with symptomatic intrathoracic cysts, including chest X-ray, CT scan, and chest MRI. All the imaging modalities showed a well-rounded mass with clear boundaries (Figure 1).

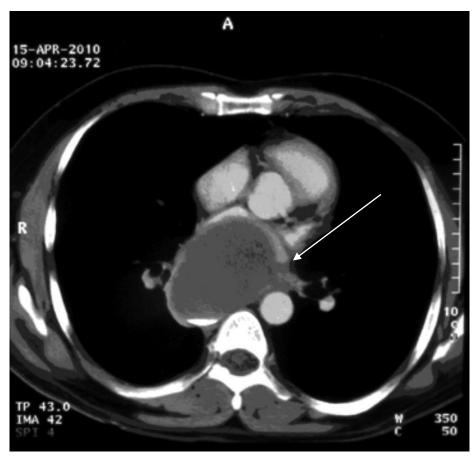


Figure 1: CT scan of a posterior mediastinal bronchogenic cyst, with a well-defined rounded mass indicated by a white arrow

After resection, the cysts were sent to the histopathology department for final diagnosis. All cysts were lined with respiratory epithelium (i.e., pseudostratified columnar epithelium) and the walls contained smooth muscle cells (n=2), cartilage (n=3), secretory glands (n=5), skeletal muscle cells

(n=1), nerve bundles (n=2). Other findings included ulceration (n=2), lymphocytic infiltrates (n=4), anthracnosis (n=2), and squamous cell metaplasia (n=1). No cysts showed evidence of malignancy.

Surgery

All 12 patients underwent total cyst resection, of

which ten underwent thoracotomy and cystectomy, and two underwent sternotomy and excision of the cyst. The longest axis of bronchogenic cysts in our study ranged from 2–11 cm (mean = 4.52cm) (Table 1). All 12 surgeries were performed smoothly, as no complications were reported during or after the operation; no cyst recurrence was reported.

DISCUSSION

A bronchogenic cyst is a rare congenital anomaly of the tracheobronchial tree, which originates from the abnormal budding of the primitive foregut during embryogenesis. They account for 10–18% of all mediastinal masses. Since most patients are asymptomatic, it is difficult to determine the true incidence of such cysts [3, 8]

Since patients can present at any age, the consultation age is highly variable. Most patients present in adulthood between the third and fourth decades of life, after the development of symptoms or incidental discovery on imaging. Symptoms that appear in pediatric patients are usually severe and life-threatening and are the most common cause of symptomatic bronchogenic cysts [2].

In most cases, bronchogenic cysts are either mediastinal or intrapulmonary. Mediastinal cysts are found in the middle and posterior mediastinum, and many sites have been described, including paratracheal, carinal, hilar, and paraesophageal areas [9, 10]. Anterior mediastinal cysts, including intrathymic cysts, are extremely rare [3]. In our study, we found four cysts in the posterior mediastinum and four in the anterior, but no middle mediastinal cysts. Most intrapulmonary cysts we found were in the lower lobe [11]. In our series, two intrapulmonary cysts were in the right lower lobe, one in the left lower lobe, and the last in the right upper lobe.

Intrathymic bronchogenic cysts are very rare and are usually clinically insignificant because they rarely produce any symptoms. Most cases are discovered incidentally during chest imaging due to other reasons. CT imaging of the chest usually shows soft tissue attenuation that is due to the cysts being filled with protein-rich fluid, which can be confused with non-invasive thymoma [3]. In our series, one patient developed a symptomatic intrathymic bronchogenic cyst while the other was asymptomatic; both underwent total surgical excision of the cyst and the diagnosis was confirmed by histopathology.

Other unusual sites have been reported in the literature, including skin and subcutaneous tissues, abdomen, pericardium, diaphragm, spine, and many

other sites [12, 13].

Most bronchogenic cysts are asymptomatic [14]. Symptoms depend on size, location, and whether a connection is established with the airway. Most intraparenchymal cysts are asymptomatic, but they are more likely to produce symptoms than mediastinal cysts [8, 11]. When symptoms appear, they are usually cough, pain, and dyspnea due to airway compression [15]. In our study, one patient had no symptoms associated with a bronchogenic cyst, which was incidentally discovered during aortic valve replacement surgery. All other 11 patients were symptomatic (91.7%) (Table 2).

Complications of bronchogenic cysts are not uncommon, especially in symptomatic patients and cysts with patent communication with the airways. Larger cysts are more likely to cause recurrent pneumonia caused by compression of adjacent structures [16]. One-third of our patients suffered from serious complications related to cysts, the most common of which was pneumonia.

Less common complications include ruptured cysts, pneumothorax, pleuritis, and severe hemoptysis [17]. The patent communication with the airway is related to a higher chance of infection and hemoptysis. Rare cases of superior vena cava syndrome, malignant transformation, and arrhythmia have been reported [2].

Chest radiography is usually the first radiologic investigation performed in patients with respiratory complaints, but it is a poor diagnostic tool. CT scan is the best imaging study used to show the size, location, morphology, contents, and compressed structures affected by mediastinal masses [18], although it is used for differential diagnosis rather than a definitive diagnosis.

MRI can be used to differentiate cystic lesions more accurately from solid lesions so that when CT scans show soft tissue-like lesions, it can differentiate mediastinal bronchogenic cysts from mediastinal neoplasia [19] (Figure 1).

All symptomatic bronchogenic cysts are treated by surgical resection [20, 21], while the treatment of asymptomatic bronchogenic cyst is still a controversial topic [22]. Many recommend surgical resection due to the risk of symptomatic transformation and malignancy [15, 19]. Kirmani et al. showed that surgical treatment in asymptomatic patients had a morbidity of 20%, while 45% of conservatively treated patients eventually developed symptoms, and the risk of malignancy was 0.7% [22]. In our series, all 11 symptomatic patients and the single asymptomatic patient underwent complete cyst resection without postoperative complications.

Thoracotomy is one of the most common surgical interventions used to treat bronchogenic cysts, but newer technologies, including video-assisted thoracoscopy (VAT), robotic surgery, and transbronchial aspiration, are becoming more common. According to Lau et al., compared with thoracotomy, thoracoscopy is associated with a shorter hospital stay, less need for intensive care, and no musculoskeletal complications, although the operation time and postoperative complications of the two groups are comparable [23].

The recurrence of the bronchogenic cyst is usually caused by incomplete removal of all cystic walls. Therefore, it is recommended that the cyst be completely resected [24]. If it is difficult to entirely remove the cyst due to its location and adherence to vital structures, electrocautery can be used to ablate the remaining cystic wall to prevent future recurrence [19]. Although the recurrence rate is still unknown because most patients are lost to follow-up, recurrence up to 25 years after resection has been reported [14]. Among our patients, there was no recurrence during the follow-up period, with our

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first case having undergone surgery in 2010. The lack of recurrence may be attributed to all of our patients having undergone total resection of the cyst.

CONCLUSION

Bronchogenic cysts, although rare, can lead to dangerous complications if left untreated. Bronchogenic cysts should always be considered among the differential diagnosis when identifying mediastinal, cervical, and thymic masses in patients. The surgical approach for resection should depend on the location of the cyst to ensure complete resection with minimal surgical complications. We recommend complete resection of all bronchogenic cysts to prevent future complications, malignant transformation, and recurrence.

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الكيسات القصبية المنصفية: العرض السريري والتشخيص ونتائج العلاج

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الملخص

تعتبر الكيسات القصبية من الأورام المنصفية النادرة التي تسببها تشوهات المعى الأمامي. على الرغم من أن الجراحة لا تزال هي الشكل النهائي للتشخيص والعلاج، إلا أنه يمكننا الوصول إلى التشخيص من خلال طرق التصوير. تهدف هذه الدراسة بأثر رجعي الشكل النهائي للتشخيص والعلاج، إلا أنه يمكننا الوصول إلى التشخيص من خلال طرق التصويل. وعلامات المرضى وأعراضهم ومضاعفات الأكياس والنهج الجراحي للاستئصال والخروج الجراحي لمرضى هذه الدراسات. هذه دراسة مستعرضة وصفية بأثر رجعي استعرضت السجلات الطبية لـ 12 مريضًا تم تشخيصهم رسميًا بالأكياس القصبية من خلال التشريح المرضي وعولجوا جراحيًا بين عامي 2010 و 2020. لقد راجعنا السجلات الطبية لجميع المرضى، بما في ذلك العمر وموقع الإصابة التكيسات والأعراض والمضاعفات وتقنيات التصوير والتنخلات الجراحية تم تضمين اثنتي عشرة حالة كيسات قصبية. ثمانية كيسات منصفية (اثنان منها كانا داخل التلامس) وأربعة أكياس داخل النسيج المتني. كان أحد المرضى بدون أعراض وكان الـ 11 الباقون من الأعراض. كانت الأعراض الأكثر شيوعًا هي ألم الصدر وضيق التنفس والسعال. 4 حالات عانت من مضاعفات حادة في كيس الشعب الهوائية، منها 3 حالات مصابة بالتهاب رئوي وحالة واحدة مصابة بانخماص الرئة. تراوح أطول محور للكيس القصبي من 2 سم إلى 11 سم (المتوسط = 4.5 سم). خضع جميع المرضى الـ 12 لاستئصال جراحي كامل للكيس دون مضاعفات أو تكرار ما بعد الجراحة. على الرغم من ندرة الأكياس بدون أعراض، يعتبر الاستئصال الجراحي الكامل هو الخيار الأفضل لمنع تكرار حدوث مضاعفات في المستقبل، مثل الورم الخبيث. الكلمات الحالة: المنصف؛ كيس قصبي المنشأ بضع الصدر.