Presentation and Management of Bronchogenic Cysts in the Adult*

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Bronchogenic cysts are congenital anomalies of the bronchial tree that are often asymptomatic at presentation in adults. Management of asymptomatic bronchogenic cyst in this population remains controversial. Eighteen patients with bronchogenic cysts were treated at our institution since 1975. At initial presentation, 10 patients (56 percent) were asymptomatic and 8 (44 percent) were symptomatic. Cough and pain were the most frequent symptoms. Two patients presented with potentially serious complications, one with respiratory distress from airway compression and the other with infection and airway fistulae. Chest radiographs were abnormal but nondiagnostic in 17 out of 18 (94 percent) patients. Chest computerized tomography (CT) scans were abnormal in eight of eight (100 percent) patients, but they confirmed the benign cystic nature in only five of eight (62.5 percent). Overall, considering the use of all imaging modalities and clinical suspicion, bronchogenic cyst was considered in the preoperative differential diagnosis in only 11 of 18 (61 percent) patients. Fifteen of 18 cysts were resected initially. Three of the asymptomatic patients who were followed up initially ultimately required resection because of the development of symptoms. A trend toward increased postoperative complications was noted in patients who were symptomatic at the time of surgery (27 percent vs 14 percent). In conclusion, adult patients with asymptomatic bronchogenic cyst may develop symptoms over time. Symptoms in adults can sometimes be potentially serious. Since a confident preoperative diagnosis is not always possible and because surgical complications may be more common in the symptomatic patient, we recommend surgical resection of all suspected bronchogenic cysts in operable candidates. (Chest 1994; 106:79-85)

CT=computerized tomography; FNA=fine needle aspiration

Key words: bronchogenic cysts, fine needle aspiration

Bronchogenic cysts represent a spectrum of bronchopulmonary malformations that result from an abnormal budding of the tracheobronchial tree. The tracheal bud develops from the primitive foregut as a ventral diverticulum around the fourth week of gestation and then undergoes further branching and differentiation. The bronchogenic cyst develops as part of the tracheobronchial tree separate from the primary airway as a result of aberrational development.

Bronchogenic cysts have a wide range of clinical and radiologic manifestations. Airway compression of the supple tracheobronchial tree in infancy and early childhood often leads to symptoms and occasionally life threatening complications. Bronchogenic cysts in the adult frequently present as an incidental radiologic finding. Some reports suggested that such asymptomatic cysts were of little clinical importance and could be followed up. Conversely, more recent studies suggest these cysts are frequently symptomatic in the adult. Surgical excision of the asymptomatic cyst is recommended to prevent complications and avoid the operative difficulty associated with resecting symptomatic cysts. Unfortunately, the true natural history of these cysts in adults is uncertain since an unknown percentage of asymptomatic adult patients with bronchogenic cysts remain undiagnosed. The available series, including our own, reflect a significant selection bias in favor of the symptomatic individual. Interpretation of the available literature is further complicated by diagnostic uncertainty in the absence of surgical resection for a definitive diagnosis. Despite improved imaging modalities, radiologic studies including computerized tomography (CT) cannot always exclude the possibility of noncystic solid tumors. Although some reports in the last decade have suggested that the presence of bronchial epithelial cells on fine needle aspirate (FNA) may be diagnostic of bronchogenic cysts, the specificity of this finding is unknown.

We present a retrospective review of bronchogenic cysts in adults treated at the Cleveland Clinic over the last 17 years.

PATIENTS AND METHODS

The medical records of all patients who underwent surgical excision of pathologically confirmed bronchogenic cysts at our institution during the period from 1975 to 1992 were reviewed. Data were collected with regard to demographics, mode of pre-

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sensation, reliability of common imaging techniques (chest radiograph, tomogram, and CT), management, surgical findings, and incidence of operative and postoperative complications stratified by symptom status at the time of surgery. To determine the diagnostic utility of bronchial epithelial cells obtained by FNA, we recorded their occurrence in FNA specimens in patients who had this procedure done in our series of confirmed bronchogenic cysts and in consecutive patients with confirmed diagnosis other than bronchogenic cysts.

**RESULTS**

During a 17-year period between 1975 and 1992, 18 adult patients underwent surgical excision of thoracic bronchogenic cysts. There were 12 men and 6 women and their ages ranged from 17 to 70 years (mean 37.6 years). The follow-up period after surgical excision ranged from 12 months to 11 years with one patient being lost to follow-up because of residence abroad.

**Clinical Presentation and Location**

Table 1 compares asymptomatic and symptomatic patients. Ten patients (56 percent) were asymptomatic at initial presentation. Their mean age was 38 years. In 9/10 (90 percent) the cyst was found incidentally on a chest radiograph done as part of a physical or pre-employment screening and in one patient it was found incidentally at cardiac surgery. Nine (90 percent) of the asymptomatic cysts were mediastinal and 1 (10 percent) was intraparenchymal in location.

Eight patients (44 percent) were symptomatic at initial presentation with a mean age of 38 years (Table 2). Cough and pain were the most common symptoms. The pain was located in the chest and back in two, chest and epigastrium in one, chest and shoulder in one and chest alone in two. One patient presented with acute respiratory distress, stridor, and hypoxemia from airway and vascular compression. Most symptomatic patients had more than one symptom.

Five (63.5 percent) of the symptomatic cysts were mediastinal and three (37.5 percent) were intraparenchymal. The mediastinal cysts were located in the anterior mediastinum, (3) in the middle mediastinum, (5) in the posterior mediastinum, (4) and in both the middle and posterior mediastinum (2). The four intraparenchymal cysts were located in the right lower lobe (2) in the right upper lobe (1), and in the left lower lobe (1). Combining both symptomatic and asymptomatic patients, 14/18 (78 percent) were mediastinal and 4/18 (22 percent) were intraparenchymal. The incidence of symptoms between mediastinal and intraparenchymal cysts did not differ significantly (Fisher's exact test, p=0.27).

**Diagnostic Techniques**

A variety of radiologic and endoscopic diagnostic tests were performed depending on initial clinical suspicion, cyst location, and decade of presentation. These included chest radiograph (18), CT scan (8), tomogram (4), digital subtraction angiography (3), barium swallow (4), thyroid scan (2), esophagoscope (3), mediastinoscopy (2), bronchoscopy (7), two dimensional echocardiography (2), transesophageal echocardiography (2) (Fig 1), and FNA (6). Table 3 shows the sensitivity and clinical use of the commonly used imaging modalities. The chest radiograph was abnormal but nondiagnostic in 17/18 (94 percent) patients (Fig 2). Chest radiographs were helpful in defining the location, size, and contour of the lesion. Based on the chest radiographic appearance, bronchogenic cyst was considered in the preoperative differential diagnosis in nine (50 percent).

**Table 1—Comparison of Asymptomatic and Symptomatic Groups**

<table>
<thead>
<tr>
<th>Group</th>
<th>Age (Mean Range)</th>
<th>Location*</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No. (%)</td>
<td>Mediastinal (No. %)</td>
</tr>
<tr>
<td>Asymptomatic</td>
<td>38 (17-70)</td>
<td>9 (90)</td>
</tr>
<tr>
<td>10 (56)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Symptomatic</td>
<td>38 (18-60)</td>
<td>5 (63.5)</td>
</tr>
<tr>
<td>8 (44)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>All patients</td>
<td>38 (17-70)</td>
<td>14 (78)</td>
</tr>
<tr>
<td>(18)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*Frequency of symptoms in mediastinal vs parenchymal was not significantly different (Fisher’s exact test, p=0.27).
The chest CT scan was abnormal in eight of eight (100 percent) patients evaluated since 1977. The CT scanning was helpful in further delineating the location, relation to adjacent structures, and differentiating the abnormality from vascular structures. Using Kuhlman's criteria29 to define a benign cyst (Fig 3), the CT scan was strongly suggestive of a benign cyst in five (62.5 percent). Bronchogenic cyst was considered in the preoperative diagnosis in seven (87.5 percent). The chest tomogram, performed before the availability of CT, was abnormal in all four patients (100 percent), but suggested a benign cyst in only one. Considering all the above imaging modalities, bronchogenic cyst was included in the preoperative differential diagnosis in 11/18 (61 percent).

Six patients in this current series had FNA. Three had a transbronchial FNA via a flexible bronchoscope and all three had bronchial epithelial cells in their specimens. By comparison, only one of three patients had bronchial epithelial cells in a transthoracic FNA specimen. One had cells suspicious for carcinoid. To further examine the diagnostic utility of finding bronchial epithelial cells on a needle aspirate, we looked at ten consecutive transbronchial FNA and ten transthoracic FNA in patients with diagnosis other than a bronchogenic cyst. Nine (90 percent) of these ten transbronchial specimens and four (40 percent) of ten consecutive transthoracic FNA specimens had bronchial epithelial cells indicating poor specificity.

Management

Seven of the eight patients who were symptomatic had early surgical resection. One patient who presented with acute respiratory distress and pulmonary artery compression (on echocardiography and CT) had the cyst urgently decompressed by mediastinoscopy allowing clinical stabilization before surgical resection. The reasons for intervention in these patients were persistent symptoms in four and diagnostic uncertainty in the other four. Diagnostic uncertainty led to early surgical excision in six of ten asymptomatic patients (one via mediastinoscopy). One additional patient had the cyst removed when it was found incidentally in cardiac surgery.

Surgical resection was required ultimately in the three remaining asymptomatic patients who developed symptoms. The three patients developed cough with increasing cyst size at 10 years, recurrent infection with development of a right bronchial fistula at 3 years, and persistent pain after FNA at 1½ years respectively.

Operative Findings

Surgical excision was performed through a standard posterolateral thoracotomy in 14, median ster-

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Table 3—Utility of Imaging Studies

<table>
<thead>
<tr>
<th>Type</th>
<th>No.</th>
<th>Abnormal (%)</th>
<th>Suggestive of Benign (%)</th>
<th>Considered in Differential Diagnosis (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Chest x-ray</td>
<td>18</td>
<td>17 (94)</td>
<td>—</td>
<td>9 (50)</td>
</tr>
<tr>
<td>CT scan</td>
<td>8</td>
<td>8 (100)</td>
<td>3 (62.5)</td>
<td>7 (87.5%)</td>
</tr>
<tr>
<td>Tomogram</td>
<td>4</td>
<td>4 (100)</td>
<td>1 (25)</td>
<td>2 (50)</td>
</tr>
</tbody>
</table>

*With use of the above imaging modalities, bronchogenic cyst was considered in the differential diagnosis in 11/18 patients (61%).

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FIGURE 2. Chest radiograph of a patient with a left lower lobe intraparenchymal bronchogenic cyst during an episode of infection (left) and following treatment of infection (right).

FIGURE 3. A CT scan of the chest in a patient with middle mediastinal cyst showing classic signs of a benign cyst (rounded, smooth walled, well demarcated with homogenous low attenuation and without vascular enhancement or infiltration of adjacent structures).
notomy in 3, and a cervical mediastinoscopy in 1. The four patients with parenchymal cysts underwent lobectomies. The size of the cyst at surgery ranged from 3 to 10 cm (mean 6 cm). Operative difficulties were encountered in 7/11 (64 percent) of patients who were symptomatic at the time of surgery. Surgical resection in these patients was complicated by marked pleural or mediastinal adhesions (3), adhesions and compression of mediastinal structures (2), extensive pericystic adhesions and cyst-airway fistula (1), and origin in the muscularis propria of the esophageal wall (1).

Although two of seven asymptomatic patients had nonpatient communications with the tracheobronchial tree, operative difficulty was described in only one patient who had a cyst in the posterior mediastinum with partial involvement of the esophageal wall. Despite a trend toward more operative difficulty occurring in the symptomatic as compared with the asymptomatic patient, this difference was not significant statistically (Fisher’s exact test, p=0.0656).

Operative and Postoperative Complications

No operative or postoperative mortality occurred. Three out of the 11 patients (27.3 percent), who were symptomatic at the time of surgery developed complications (Table 4). One patient with a large middle mediastinal cyst and extensive pericystic adhesions developed a phrenic nerve paresis. The patient with esophageal involvement who required an intercostal muscle patch of the esophageal bed subsequently developed an esophageal stricture and ossified esophageal wall. He ultimately required esophagectomy 10 years after the initial surgery. A third patient who initially had surgery at an outside hospital for an infected cyst with cyst airway fistula requiring resection and closure of the fistula, developed recurrence of the cyst, infection, and two additional bronchial fistulae (Fig 4). The excision of the cyst required a partial pericardectomy, sleeve resection of the right mainstem bronchus, and pleuropericardial fat wrapping of the bronchial anastomosis. The recurrence was secondary to incomplete cyst resection at the original hospital because it could not be excised in toto due to extensive pericystic adhesions.

Only one of seven (14 percent) patients who were asymptomatic at the time of surgery developed complications. This consisted of a persistent pneumothorax after resection of a right upper lobe cyst requiring prolonged chest tube drainage for 14 days. The difference in complications between the symptomatic and asymptomatic patients was not significant (Fisher’s exact test p=0.596). Overall 16/18 patients in this study have been symptom free after surgery with follow-up ranging from 1 to 11 years. Two patients who developed delayed complications in the form of an esophageal stricture and a recurrent cyst are also symptom free at 2 years and 5 years respectively after treatment of their complication.

**DISCUSSION**

Bronchogenic cysts are one of the most common bronchopulmonary malformations. Though more commonly mediastinal in location, cysts arising at a later stage of development may be intraparenchymal and in rare cases have occurred in prestenral tissues, diaphragm, spine, skin and subcutaneous tissue, pericardium, neck, abdomen, or have extended from the mediastinum through the diaphragm into the abdomen—“dumbbell” cysts. The cyst wall is lined by ciliated pseudostratified columnar epithelium and often contains bronchial mucus glands, smooth muscle, and cartilage.

Since the first reported case of bronchogenic cyst by Meyer in 1859, there have been several reports and literature reviews in both children and

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**Table 4—Operative and Postoperative Complications**

<table>
<thead>
<tr>
<th>Symptom Status at Surgery (No.)</th>
<th>Complication Type</th>
<th>Type of Complication</th>
</tr>
</thead>
<tbody>
<tr>
<td>Asymptomatic (7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Prolonged air leak</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Right phrenic nerve paresis</td>
<td></td>
</tr>
<tr>
<td>Symptomatic (11)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Esophageal mucosal laceration followed by esophageal stricture</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Recurrent cyst with right mainstem fistula</td>
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</tr>
</tbody>
</table>

*Frequency of complications in patients symptomatic vs asymptomatic at time of surgery was not statistically significant (Fisher’s exact test p=0.596)
Management of the asymptomatic cyst in the adult remains controversial, with some authors suggesting continued observation.4-7 Fontenelle et al14 in a study of young military recruits who had routine chest roentgenograms, reported that 77 percent of patients with bronchogenic cysts were asymptomatic. More recent series,15-22 however, suggest that the majority of adults with bronchogenic cysts eventually develop symptoms. In the largest reported series of 86 patients, St. Georges et al22 noted that with prolonged observation, 72 percent of patients became symptomatic. In our series, all three patients who were asymptomatic initially and followed up, subsequently developed symptoms.

The most common symptoms are from compression of mediastinal structures, infection, or hemorrhage within the cyst.5 Other serious reported complications in adults include extrinsic pulmonary artery stenosis,39 superior vena caval obstruction,41 pericardial tamponade,42 arrhythmias,43 unilateral pulmonary edema,10 obstructive emphysema,44 bronchial atresia,45 hypoplastic pulmonary artery,46 pneumonia,47 unilateral ventilation perfusion defect,48 pleural effusion,49 thrombosis,50 an air embolism that occurred during rapid decompression of the patient after working in an underground tunnel,51 severe hemoptysis,20 and carcinomatous52,53 or sarcomatous change.54 Two patients with mediastinal cysts in our series presented with potentially life-threatening complications. One patient arrived with acute respiratory distress from airway compression and the other presented with signs of infection and right mainstem bronchial fistulae.

A variety of conditions are considered in the differential diagnosis of both parenchymal and mediastinal bronchogenic cysts. With parenchymal cysts, the differential diagnosis includes neoplasms, granulomas, hematomas, vascular malformation, lung sequestration, lung abscesses, infected bullae, and hydatid cysts.55 The differential diagnosis of mediastinal bronchogenic cysts includes metastatic tumor, lymphoma, teratoma, neurogenic tumors, embryonal sarcoma, inflammatory lymphadenopathy, pulmonary sequestrations, hemangioma, lipoma, and neuro-enteric, pericardial, or esophageal duplication cysts.21 Classically, the chest radiograph reveals a rounded, well-demarcated, noncalcified mass with a homogenous “water density.”23 Unusual radiologic features caused by air trapping, infection, or rupture of the cyst with development of an air fluid level, a lobulated contour, or variations in the density of the cyst caused by calcification, surrounding atelectasis or pneumonia may contribute to diagnostic uncertainty.21 The limitations of the chest radiograph were underscored in this series where a bronchogenic cyst was considered in the differential diagnosis in only 50 percent of cases despite the presence of a radiographic abnormality in 94 percent. Similarly, in a review of multiple series by Haddon and Bower,25 the possibility of a bronchogenic cyst on the basis of the chest radiograph alone was considered in only 20 percent to 77 percent of the patients.

The CT scan provides better definition of the cyst and has eliminated virtually the need for other radiographic studies.21,23,36 Classically bronchogenic cysts are of water density (0-20 Hounsfield units); however, in the presence of infection or in those with a variable protein and calcium content, the density may be higher, falling into the solid tissue range23-25 and thereby increasing the diagnostic uncertainty. In this series, the CT scan was abnormal in all cases (eight of eight, 100 percent), but suggested a classic benign cyst in only 50 percent of the cases. Based on the CT appearance, however, bronchogenic cyst was considered in the differential diagnosis in 87.5 percent of the cases. Combining all imaging modalities, bronchogenic cyst was considered in the preoperative diagnostic differential in 61 percent of our patients.

In the series reported by Cartmill et al,21 a preoperative diagnosis was reached on radiologic grounds in only 9/20 patients (45 percent) the increased use of CT scans has fueled the management debate. Pugatch et al16 followed up 4 asymptomatic patients who met CT criteria for a benign cyst for a period ranging from 6 months to 3 years.16 No patient developed symptoms and the authors suggested similar asymptomatic patients could be followed up conservatively. This conclusion is not supported by St. Georges et al22 where 24/37 (65 percent) of the asymptomatic patients required surgery when the bronchogenic cyst enlarged or became symptomatic over a period of 6 months to several years. In our series, all 3 asymptomatic patients who were followed up clinically for ½ years to 10 years eventually required surgery because of the development of symptoms or cyst enlargement. Both series suggest that the asymptomatic patient remains at risk for the development of symptoms and complications. Management recommendations cannot be based on short-term follow-up.

The diagnosis of thoracic masses has been advanced by the introduction of FNA (performed either via the bronchoscope or percutaneous route).56 In 1981, Delarue et al26 were the first to suggest that the presence of bronchial epithelial cells on FNA may be pathognomonic for bronchogenic cysts, but details of the technique or route were not clarified.26 Based on a review of two cases, Schwartz et al27 in 1985 reported that transbronchial FNA can be used to diagnose bronchogenic cysts if bronchial epithelial cells in mucus are observed. Thereafter, in 1986 Schwartz et al,28 based on a single case report, suggested that
transbronchial FNA is a safe technique that can be used not only for diagnosis but also for managing bronchogenic cyst. The diagnostic value of the presence of bronchial epithelial cells on FNA, however, remains unclear since no studies have reported the sensitivity or specificity of such a finding. In our series, four of six (67 percent) of the needle aspirates were positive for bronchial epithelial cells (three transbronchial and one percutaneous). Transbronchial FNA in patients with mediastinal disease from other causes, however, reveals that bronchial epithelial cells are found routinely regardless of the underlying diagnosis. Similarly, bronchial epithelial cells were also identified in percutaneous thoracic FNA specimens. Hence, we do not consider the finding of bronchial epithelial cells on FNA either via the transbronchial or transthoracic route a diagnostic end point.

Transbronchial or transesophageal needle aspiration of cysts meeting classic criteria for a bronchogenic cyst has been recommended as a practical alternative to surgery in the management of mediastinal cysts. However, the reported follow-up without recurrence has only ranged from 3 months to a year.29 FNA is useful in decompression of cysts in acutely compromised or nonoperative candidates,37 but may be associated with recurrence of the cyst.38 Definitive treatment remains complete surgical excision.

Surgical excision usually is via a posterolateral thoracotomy or median-sternotomy,19,36 with intrapulmonary cysts requiring segmental or lobectomy. Most cysts can be excised surgically with minimal morbidity,21 but at times, because of dense pericystic adhesions to adjacent structures or large cyst size, surgical excision may be difficult and lead to only partial excision or complications.22 In the series reported by St. Georges et al.,22 major operative difficulties or intraoperative complications were encountered in 29/86 (44 percent) of the patients, all of whom were symptomatic preoperatively. In our series, a trend toward greater operative difficulty and more operative and postoperative complications occurred in the patients symptomatic at the time of surgery. Recurrence of bronchogenic cysts has been reported after incomplete surgical removal,39 as was seen in one patient in this series with an infected fistulous mediastinal cyst and airway fistulae that had initially been incompletely excised at another hospital.

Other suggested alternatives to surgical excision via thoracotomy are transbronchial needle aspiration,28,29 transesophageal needle aspiration,60 percutaneous aspiration,61 transtracheal aspiration,62 extrapleural aspiration,63 removal via mediastinoscope64 (as was done in one patient in this series), thoracoscopic resection,61 and imaged (video assisted) thoracoscopy resection.65 Since aspiration is not recommended as a permanent therapeutic modality given the likelihood of cyst recurrence and mediastinoscopic resection is possible in only an occasional case, video-assisted thoracoscopic resection is perhaps the only technique that holds promise as a definitive management option to resection via thoracotomy.65

In summary, based on our series and literature review, we can conclude that in adults (1) asymptomatic bronchogenic cysts may become symptomatic with time; (2) bronchogenic cysts may be associated with potentially serious complications; and (3) since a confident preoperative diagnosis is not always possible and resection of symptomatic cysts may be associated with greater operative difficulty and more operative/postoperative complications, we favor excision of all suspected bronchogenic cysts in operable candidates.

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Presentation and Management of Bronchogenic Cysts (Patel et al)


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