CASE REPORTS

Resection of 8 Mediastinal Bronchogenic Cysts by Video-Assisted Thoracoscopy

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Mediastinal bronchogenic cysts are an uncommon entity and surgical experience of their removal by video-assisted thoracoscopy is limited. We present our patient outcomes and surgical technique in the treatment of bronchogenic cysts by video-assisted thoracoscopy. The study included 8 patients (4 females and 4 males between the ages of 4 and 52 years), 7 of whom presented clinical symptoms. The mean widest diameter of the cyst was 7.6 cm. In 5 patients the cyst was in the middle mediastinum and in 3, the posterior mediastinum. The intervention was performed using 3 or 4 entry points. Initial puncture of the cyst and removal of its contents greatly facilitated cyst manipulation and subsequent dissection of the cyst sac from the structures to which it was attached. In all 8 cases resection by video-assisted thoracoscopy was carried out with no intraoperative complications. The mean postoperative hospital stay was 3.3 days. During follow-up, which ranged from 4 months to 10 years, no patients presented late-onset or recurrent complications.

Key words: Mediastinal tumor. Mediastinal cyst. Bronchogenic cyst. Thoracoscopy. Video-assisted thoracoscopy (VATS).

Introduction

Bronchogenic cysts account for 18% of all primary mediastinal tumors and are the most common cystic lesions in this anatomical location.1,2 These congenital abnormalities of the tracheobronchial tree may develop in the pulmonary or the mediastinal parenchyma depending on where in relation to the branch from the primary airway they are located, although atypical sites have been reported.

Bronchogenic cysts are lined with a ciliated columnar epithelium, contain cartilage, and are filled with mucoid material. They are usually unilocular although they may contain internal trabeculation. They are often asymptomatic and the symptoms depend on cyst location or complications, such as intracystic hemorrhage, rupture, infection, or metaplastic changes.3-6

Magnetic resonance imaging tends to be the current imaging modality of choice for establishing diagnosis and determining cyst location, but computed tomography and endoscopic ultrasound are also useful—although distinguishing between bronchogenic cysts and other mediastinal tumors is occasionally difficult.7,8 Standard surgical treatment is removal of the cyst by thoracotomy.9

Since the development of video-assisted thoracoscopic surgery, encouraging outcomes using this approach have been reported in isolated cases and in small series.9-13

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Case Descriptions

In our hospital, from June 1994 through December 2006, 8 patients diagnosed with bronchogenic cyst underwent video-assisted thoracoscopic surgery (Table). Six patients presented clinical symptoms (the presenting complaint was cough in 4 patients, dyspnea in 1, and hemoptysis in 1), the cyst of another patient was a chance finding, and the remaining patient had a ruptured cyst in the left pleural space that manifest as a hydropneumothorax. In all the 8 cases, preoperative diagnostic studies included simple chest x-ray, computed tomography, and/or magnetic resonance imaging (Figures 1A and 1B). Esophagography was also performed in 2 cases. Mean cyst diameter was 7.5 cm (range, 5-10 cm). According to the Maier classification,14 the cyst was located in the middle mediastinum in 5 cases (1 subcarinal and 4 paratracheal) and in the posterior mediastinum in 3 cases.

Surgery was carried out with patients in lateral decubitus, positioned for posterolateral thoracotomy. The video camera was passed through the first entry point at the level of the seventh or eighth intercostal space in the midaxillary line. The position of the remaining entry points depended on the location of the cyst; in most cases the sites were at the sixth space in the postaxillary line, at the third space in the anterior axillary line, and at the fifth space in the anterior axillary line. First, a complete examination of the pleural cavity was carried out and it was determined whether the cyst was anatomically connected to adjacent structures, especially the vena cava and the esophagus. In the first 2 cases dissection began with the cyst closed, but accidental rupture and subsequent evacuation greatly facilitated evacuation of cyst contents by aspiration.

Characteristics of Patients With Mediastinal Bronchogenic Cysts

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex</th>
<th>Age, y</th>
<th>Location</th>
<th>Diameter, cm</th>
<th>Symptoms</th>
<th>Complications</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>14</td>
<td>Middle (paratracheal)</td>
<td>7</td>
<td>Cough</td>
<td>No</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>27</td>
<td>Posterior</td>
<td>8</td>
<td>Cough</td>
<td>Pneumothorax</td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>4</td>
<td>Middle (paratracheal)</td>
<td>8</td>
<td>Cough</td>
<td>No</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>37</td>
<td>Posterior</td>
<td>5</td>
<td>Dyspnea</td>
<td>No</td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>45</td>
<td>Posterior</td>
<td>10</td>
<td>Cyst rupture</td>
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</tr>
<tr>
<td>6</td>
<td>F</td>
<td>52</td>
<td>Middle (paratracheal)</td>
<td>10</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>48</td>
<td>Middle (subcarinal)</td>
<td>5</td>
<td>Hemoptysis</td>
<td>No</td>
</tr>
<tr>
<td>8</td>
<td>M</td>
<td>21</td>
<td>Middle (paratracheal)</td>
<td>7</td>
<td>Cough</td>
<td>No</td>
</tr>
</tbody>
</table>

Abbreviations: F, female; M, male.

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Figure 1. Computed tomography scan of the chest (A) showing a multilocular cystic lesion located in the posterior mediastinum, and magnetic resonance image (B) showing a mediastinal cystic lesion with high signal intensity, indicating a bronchogenic cyst.

Figure 2. Video-assisted thoracoscopic images showing the entire paratracheal bronchogenic cyst and the initiation of evacuation of cyst contents by aspiration.
dissection and removal. In the remaining 6 cases, we initiated the procedure by opening the cyst and evacuating its contents, first separating the mediastinal pleura enveloping the cyst and then making a small incision in the upper part of the cyst through which to pass an aspirator to remove its content (Figure 2). This maneuver made it easier to pull the cystic sac to one side or the other and helped in the separation from adhering structures, especially the vena cava superior. The procedure continued with complete removal of the cyst using endoscissors and electrocoagulation—except in the last intervention, performed with an ultrasonic scalpel, which had previously been unavailable to us. Lung re-expansion was verified and absence of both parenchymatous and bronchial or tracheal air leaks was confirmed by the lack of rising air bubbles on immersing the parenchyma in a saline solution while the lung was insufflated. A low posteriorly-directed chest tube was then positioned for 48 to 72 hours.

In cases of cysts in the posterior mediastinum—as in those of esophageal duplication cysts—intraoperative esophagoscopy was helpful in monitoring the integrity of the mucosa. In 1 of our patients, endoscopic ultrasound revealed contact between the cyst and the esophageal wall, and a fragment of affected esophagus muscle had to be removed along with the cyst. During dissection, the possibility of air leaks was monitored by intraoperative esophagoscopy. The procedure involved instilling saline solution and insufflating air into the lungs. The lack of bubbles confirmed the absence of air leaks and hence the integrity of the mucosa.

No intraoperative complications occurred. In one patient, a follow-up chest x-ray performed at his first check-up at 15 days revealed a small right pneumothorax that had not been observed at discharge. It resolved with respiratory physiotherapy. In case 6, a routine radiographic examination at 12 months revealed an image similar to that observed before surgery and computed tomography confirmed the presence of fluid content. Recurrence was suspected and another video-assisted thoracoscopy was performed using the same entry points. In precisely the same site where the resected cyst had been, lung adhesions were observed on the mediastinal pleura. Release of the adhesions revealed a cavity containing a small amount of pleural fluid. According to the histological study, the cavity was formed from pleural wall with no respiratory epithelium or bronchial tissue. Two years after the second intervention the patient remained asymptomatic and presented no evidence of recurrence.

Mean postoperative hospital stay was 3.3 days (range, 2-5 days). During follow-up, which ranged from 4 months to 10 years, no patient presented late-onset or recurrent complications.

Discussion

Bronchogenic cysts are relatively uncommon, and most groups report low incidences. Our series of 8 patients, all of whom underwent resection by video-assisted thoracoscopy, is the largest series reported by a single hospital in Spain.

Bronchogenic cysts are benign tumors and, although they are sometimes chance radiologic findings, they can cause a wide variety of symptoms and complications. Some authors report that bronchogenic cysts in adults are often symptomatic and that a high proportion of initially asymptomatic cases eventually present symptoms. Cough and chest pain, the most common symptoms, are usually caused by compression of adjacent structures. Seven of our patients presented symptoms—cough being the predominant one.

Preoperative diagnoses of bronchogenic cysts are usually based on routine chest x-rays, which often show a well-defined, homogeneous, roughly spherical mass in the mediastinum. Computed tomography or magnetic resonance imaging needs to be performed in order to complete the evaluation. Although both imaging techniques are valid, at present magnetic resonance imaging provides better cyst definition and visualization of anatomical connections, showing low signal intensity in T1-weighted images and high signal intensity in T2-weighted images. Five of our patients (83.3%) who underwent computed tomography received diagnoses based on such images; the sixth case required magnetic resonance imaging to resolve uncertainty; the seventh and eighth underwent magnetic resonance imaging merely to confirm diagnosis.

Regarding therapeutic approach, Bolton and Shahian recommended surgical treatment only for symptomatic patients. However, St Georges et al recommended resection in all cases after observing their series of 86 patients over prolonged follow-up and noting that 72% developed symptoms and/or complications. Findings from larger series suggest that resection should be performed in all patients in view of the following: a) a confirmed diagnosis can only be made after the pathologist’s assessment, b) a large proportion of patients become symptomatic or present complications that impede treatment, and c) malignant transformation is a possibility, albeit a rare one.

Routine surgical procedure has traditionally been complete cyst removal by thoracotomy, despite reports of recurrence 25 years after surgery. Some authors have pointed out that if complete resection is impossible and remnants of cyst wall may remain in the patient, electrocoagulation of the mucosa can prevent recurrence. Accumulated experience with minimally invasive surgery has enabled a less traumatic approach in the treatment of mediastinal lesions. Since 1991, when Mouroux et al reported the removal of a posterior mediastinal bronchogenic cyst by video-assisted thoracoscopy, other groups have reported their experience using this surgical technique. Recently Yoshino et al performed resection of a posterior mediastinal bronchogenic cyst using the da Vinci robotic system.

Hazeldigg et al reported favorable outcomes using video-assisted thoracoscopy in a series of 7 bronchogenic cysts. In only a single case was removal incomplete; no intraoperative or postoperative complications were recorded, and no recurrence was observed after a mean of 6 months of follow-up. In a report by Martinod et al on a French multicenter series of 20 mediastinal bronchogenic cysts treated by thoracoscopic surgery in adult patients, thoracotomy was required in 7 patients (35%); in 5 patients resection was impossible owing to adherences, and in 2 patients thoracotomy was necessary owing to intraoperative hemorrhage. In children, Michel et al had to perform thoracotomy to complete resection in 3 cases (20%), and in 3 other cases they left a fragment of wall shared between the cyst and the bronchus. Two of our patients were younger than 15 years of age, and the remaining 6 were adults. No thoracotomies were needed and in all cases complete resection was performed by video-assisted thoracoscopy.
with similar outcomes to those obtained by Hazelrigg et al. Unlike Weber et al., who dissected the cysts and then evacuated cyst content prior to removal, we carried out complete aspiration of mucoidal liquid at the beginning of the intervention, thereby facilitating dissection.

In conclusion, we believe that mediastinal bronchogenic cysts should be treated surgically. Factors supporting our position are the high proportion of symptomatic patients in our series, the possibility of complications, and the difficulty of intervention after inflammatory phenomena eventually arise. Observation should be reserved for those patients who refuse surgical intervention or those whose condition renders them unfit for undergoing surgery. In fact, video-assisted thoracoscopic surgery should be the first approach attempted. While difficulties in removal and possible complications and recurrence are similar to those of classical posterolateral thoracotomy, video-assisted thoracoscopic surgery offers clear postoperative advantages.

REFERENCES