ABSTRACT

Background: Bronchogenic cysts are not uncommon in either children or adults. In children, they comprise approximately 6% of all mediastinal masses. Their presentation can range from an asymptomatic incidental finding to sudden respiratory distress.

Case Report: Video-assisted thoracoscopy was utilized to remove a bronchogenic cyst that was densely adherent to the adjacent esophagus in a child. This was accomplished with a Harmonic scalpel. The chest tube was removed on postoperative day 1, and the patient was discharged on postoperative day 2. An esophagogram obtained 2 weeks after surgery was normal, and the patient’s preoperative symptoms had not returned.

Conclusions: Bronchogenic cysts should be considered in the differential diagnoses for mediastinal masses at any age. Given their benign nature, thoracoscopy offers an excellent alternative to open thoracotomy for their removal.

Key Words: Bronchogenic cysts, Video-assisted thoracoscopy, Mediastinal masses, Mediastinal cysts.

INTRODUCTION

Bronchogenic cysts are often asymptomatic and discovered as an incidental finding. They may become symptomatic due to esophageal compression as they increase in size or from development of infection. We report the case of a 9-year-old female with a symptomatic bronchogenic cyst, undiagnosed since the age of 2 years, who underwent successful thoracoscopic excision.

CASE REPORT

A 9-year-old girl was first seen at the age of 2 years for cough and fever in the emergency room. A chest x-ray was obtained at that time, which was interpreted as normal. Nonetheless, the patient was diagnosed with pneumonia, given a prescription for antibiotics, and released. Subsequent to this, she presented several more times each year with similar complaints and with identical posteroanterior (PA) chest x-ray findings, resulting in several hospital admissions. She had no history of stridor, dysphagia, hemoptysis, or chest pain. Her only consistent complaint was one of a chronic, nonproductive cough. Her examination revealed no abnormalities, and the laboratory work was within normal limits at the time of her presentation to the pediatric surgery service.

Repeat chest x-rays were obtained (Figure 1) as well as a computed tomography (CT) scan of the chest (Figure 2). It revealed a well-circumscribed cyst in the right chest measuring approximately 3x4 cm directly adjacent to the esophagus in the midthoracic region. It was unilocular and homogeneous and did not appear to communicate with the esophagus or any neural structures. No air fluid level was present in the cyst. No mediastinal adenopathy was seen. Given these findings, the diagnosis of either an esophageal duplication cyst or bronchogenic cyst was entertained.

A thoracoscopic approach was utilized to evaluate and remove the mass (Figure 3). The medial aspect of the cyst was intimately involved with the muscular wall of the esophagus for several centimeters. A Harmonic scalpel was used to separate the cyst, which was filled with a white mucoid material, from the esophagus. A small portion of the cyst wall was left in place to avoid
perforation of the esophagus. The remaining cyst wall was de-epithelialized. Intraoperative esophagoscopy revealed no esophageal injury. A chest tube was placed at the time of surgery. The chest tube drainage was negligible, and it was removed on postoperative day one. The patient was discharged home the following day, after an uneventful recovery. She was seen at follow-up 2 weeks after surgery where a normal barium esophagram (Figure 4) was obtained, and her preoperative symptoms were completely resolved. Final pathology revealed a bronchogenic cyst without signs of malignant degeneration or infection.

**DISCUSSION**

Bronchogenic cysts are embryological remnants that are isolated from the normal development of the foregut. On rare occasions, they can be formed from tracheal diverticuli. They are almost always extrapulmonary and medial in location. They may be found incidentally on a radiographic study or once symptoms are produced as a result of compression or infection. If communication with the airways occurs, an air-fluid level is usually present. Histologically, they are lined with ciliated columnar epithelium and can contain cartilage and bronchial glands. Accepted treatment is excision because of concern about continued growth and subsequent compression of surrounding structures, the risk of infection, and the unlikely but reported possibility of malignant degeneration. Bronchogenic cysts comprise approximately 6% of all mediastinal masses in children, while foregut cysts as a group account for 15%. They have been found to account for 18% of all mediastinal tumors in adults.
Traditionally, open thoracotomy has been the method by which these cysts have been removed. Even with increasingly sophisticated radiographic and percutaneous techniques, an exact diagnosis is often uncertain, and the unequivocal diagnosis must be made surgically. Thoracoscopy offers both a minimally invasive diagnostic modality and also a highly successful method of definitive therapy for patients with bronchogenic cysts. Although many case reports exist regarding bronchogenic cysts and their minimally invasive treatments, reports of precious few series have been published regarding their thoracoscopic excision.

In 1993, Hazelrigg published a series of seven bronchogenic cysts removed thoracoscopically with favorable results. Martinod et al reported a series of 20 cases of bronchogenic cysts in adults, of which they successfully removed 13 thoracoscopically. Their reasons for conversion were bleeding in 2 cases and dense adhesions to surrounding vital structures in 5. In 5 cases, a portion of the cyst wall was left behind and the mucosal lining obliterated. No recurrences were reported. In a report by Michel et al, thoracoscopy was performed on 22 children, 15 of which were found to have bronchogenic

Figure 2. Computed tomography scan of chest at 9 years old. The bronchogenic cyst is circled.

Figure 3. Intraoperative picture (A). Removal of cyst (B).

Figure 4. Postoperative esophagogram.
cysts. Of these, 3 were converted to thoracotomy. Overall, 86% of cases were treated successfully with thoracoscopy. No recurrences were reported. In that article, the issue of incomplete excision and recurrence is raised, however. Merry et al7 reported in their series of 8 foregut duplications 1 recurrence with incomplete excision 1 year after surgery. Two other reports of recurrence have been reported,8,9 but so have several reporting successful obliteration of the mucosal lining with good results.9,10 Reported complications in these articles include the following: bronchial/tracheal laceration (both repaired endoscopically),5,7 recurrence,7 esophageal injury,6 pneumothorax,5,6 vascular injury.7 No deaths were reported.

CONCLUSION

Bronchogenic cysts are not uncommon and, as this case illustrates, can be easily misdiagnosed. Subtle findings, as on this patient’s chest x-ray taken at 2 years of age, are overlooked if one fails to consider cystic mediastinal masses in the differential diagnosis. Preoperative CT scan is imperative in planning an operative strategy.

We assert that thoracoscopy is a safe and effective method to deal with bronchogenic cysts and involves considerably less morbidity than thoracotomy does. As experience with thoracoscopy improves, so too will its acceptance as a superior alternative to thoracotomy in most cases of bronchogenic cysts.

References:
1. Hebra A, Othersen HB, Tagge EP. Bronchopulmonary malformations. In: Ashcroft KW, Holden TM, eds. Pediatric Surgery. Philadelphia: WB Saunders; 2000:273-286.
2. Rowe MI, O’Neill JA, Grosfeld JL. Mediastinal masses. In: Baxter SH, ed. Essentials of Pediatric Surgery. St. Louis: Mosby; 1995:306-310.
3. Wychulis AR, Payne WS, Clagett OT, Wollner LB. Surgical treatment of mediastinal tumors. A 40-year experience. J Thorac Cardiovasc Surg. 1971;62:379-392.
4. Hazelrigg SR, Landreneau RF, Mack MJ, et al. Thoracoscopic resection of mediastinal cysts. Ann Thorac Surg. 1993;56:659-660.
5. Martinod E, Pons F, Azorin J, et al. Thoracoscopic excision of mediastinal bronchogenic cysts: results in 20 cases. Ann Thorac Surg. 2000;69:1525-1528.
6. Michel JL, Revillon Y, Montupet P, et al. Thoracoscopic treatment of mediastinal cysts in children. J Pediatr Surg. 1998;33:1745-1748.
7. Merry C, Spurbeck W, Lobe TE. Resection of foregut-derived duplications by minimal-access surgery. Pediatr Surg Int. 1999;15:224-226.
8. Gharagozloo F, Dausmann MJ, McReynolds SD, et al. Recurrent bronchogenic pseudocyst 24 years after incomplete excision. Chest. 1995;108:880-883.
9. Read CA, Noront M, Carangelo R, et al. Recurrent bronchogenic cyst: an argument for complete surgical excision. Arch Surg. 1991;126:1306-1308.
10. Ribet ME, Copin MC, Gosselin B. Bronchogenic cysts of the mediastinum. J Thorac Cardiovasc Surg. 1995;109:1003-1010.
11. Lewis RJ, Caccavale RJ, Sisler GE. Imaged thoracoscopic surgery: a new thoracic technique for resection of mediastinal cysts. Ann Thorac Surg. 1992;53:318-320.

Presented as a case report at the Southwest Surgical Congress, San Diego, California, April 6-10, 2002.

Disclosure: Neither author has any financial interest in any device, equipment, instrument, or drug referred to in this article. No financial support was received for this article, nor do any conflicts of interest exist. No off-label use of any product was discussed.