Unusual presentation of dysphagia caused by bronchogenic cyst

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This is a brief case report about an unusual presentation of dysphagia caused by compression of a bronchogenic cyst and successful treatment by thoracoscopic resection.

Introduction

Bronchogenic cysts in the mediastinum are a rare condition. Progressive enlargement of the cyst may cause compression of adjacent organs. Dysphagia caused by bronchogenic cyst compression is quite rare. When a submucosal lesion is seen in panendoscopy, it should be included in one of the differential diagnoses. A complete resection, either by an open thoracotomy or thoracoscopic method, is usually curative. We report a patient who was successfully treated by thoracoscopic resection.

Case report

A 47-year-old man was previously healthy, with no history of trauma, pneumonia, travel or alimentary tract surgery. Two months prior to admission, the man presented with progressive dysphagia and weight loss of 11 lbs. Initially, the patient could swallow solid food but with time, the patient felt that solid food could not pass through the oesophagus. He could ingest soft food and drink without any problem. He was later admitted to our hospital for evaluation.

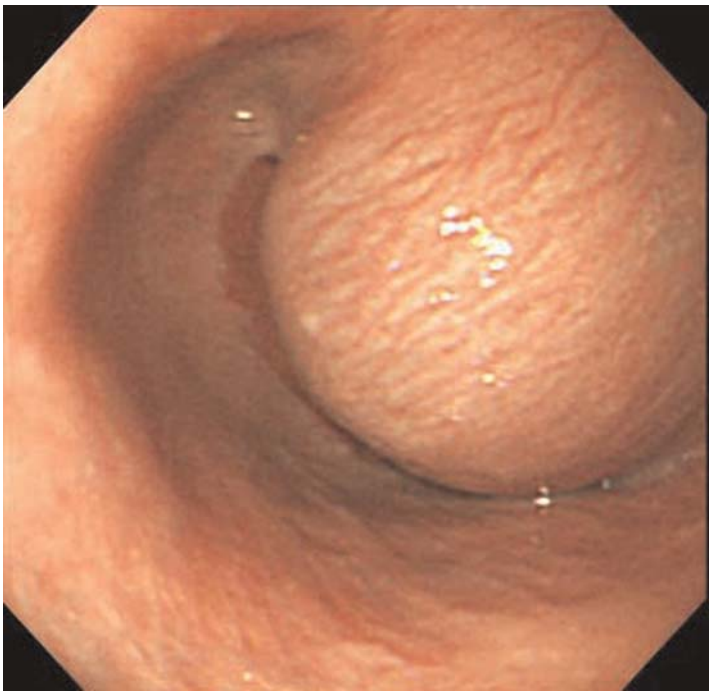
The chest radiograph taken on admission day was normal. Panendoscopy revealed one ovoid swelling with intact mucosa at 35 cm from the incisors, which was suggestive of one submucosal tumor (Figure 1). Oesophagogram also demonstrated an area of filling defect in the lower third of the oesophagus without any fistula. A chest CT scan showed a cystic lesion in the posterior mediastinum measuring around 5×5 cm adjacent to the lower third of the oesophagus without evidences of malignant features (Figures 2a and 2b).

The junction between the cyst and the oesophagus was not clear, suggesting that the cyst was probably embedded in the esophageal wall. The cystic lesion was thought to be a foregut cyst. The patient's laboratory data were all within normal limits, including complete cell counts, biochemistry, C-reactive protein, CEA, SCC, CA125 and CA19-9.

The patient underwent a thoracoscopic excision of the cystic lesion through the right side of the thorax using three thoracoports. Under thoracoscopic view, the lesion was identified beneath the mediastinal pleural after retraction of the diaphragm by a Foerster sponge forceps and division of the pulmonary ligament by way of electrical cauterization. The lung tissues were not adhered to the pleura where the cyst was located. The mediastinal pleura was opened by an L-shaped hook electrode and the capsule of the cystic lesion was identified (Figure 3a). With careful and delicate dissection by a suction tube and an L-shaped hook electrode, the cystic lesion was completely excised (Figure 3b).

During and after the procedure, the oesophageal mucosa was still intact confirmed by an oesophagoscope. The opened mediastinal pleura were repaired by interrupted sutures after removal of the lesion. Grossly, the cyst had a thin, firm and intact capsule. After being cut by a scalpel, the

Figure 1
A bulging mass in 35 cm from the incisors with near total obstruction is observed from panendoscopy



cyst had a smooth internal surface, and inside, some brown and sticky fluids were observed. Twenty-four hours after the operation, the patient started to drink and eat normally. The chest tube was removed 48 h after the operation. He was discharged on the third postoperative day. There were no pathogens cultured from the fluid. Pathological sections showed that the cyst was lined with ciliated respiratory epithelium, indicating the cyst was a bronchogenic cyst. Malignant cells were not detected in the wall and fluid by histopathological exams. Six months after surgery, he had no evidence of recurrence.

Discussion

Bronchogenic cysts are rare abnormalities and usually arise from the ventral portion of the primitive foregut. During the period of embryogenesis, the primitive foregut develops into respiratory and alimentary systems in the fourth week. Initially both the lining cells in the oesophagus and the trachea are ciliated columnar before separation of the tracheoesophageal fold. After formation of the tracheoesophageal septum, the oesophagus rapidly elongates. Any abnormality in this stage of embryogenesis may lead to malformation of the oesophagus and the trachea, which could explain why respiratory epithelium can be found in the oesophagus.[1]

Accurate preoperative diagnosis can be sometimes difficult. Ribet *et al.* reviewed 14 adults with bronchogenic cysts and found that the

Figure 2
a. A cystic lesion with a thin and slightly hyperdense capsure is shown. The junction between the cyst and the oesophagus is not clear and the lumen of the oesophagus is partially obstructed; b. The cystic lesion extends to EG junction

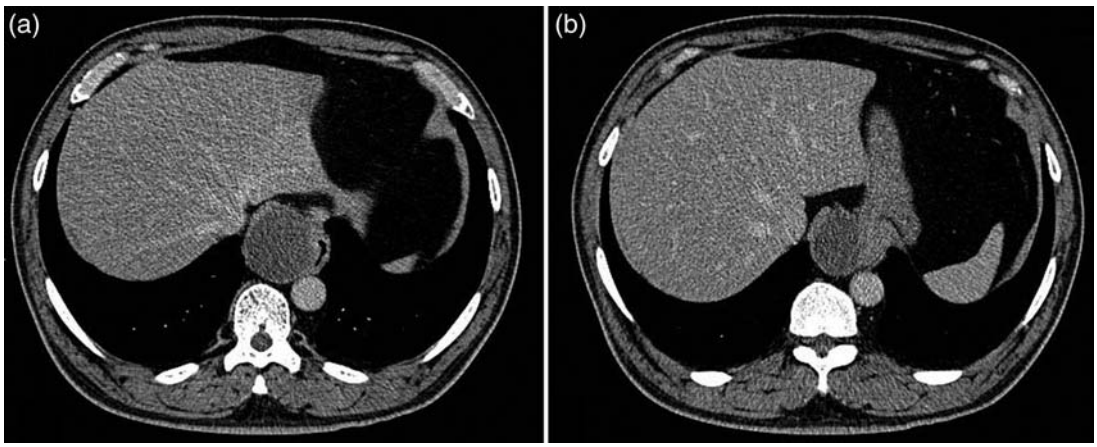
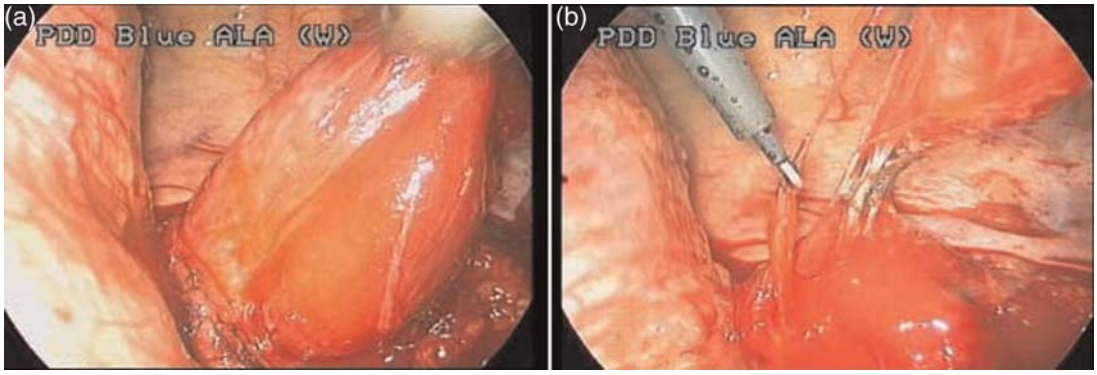


Figure 3
a. After dissection of the mediastinal pleura, the cyst is exposed; b. By both blunt and sharp dissection, the cyst is removed without entering into the oesophageal lumen



preoperative diagnoses were incorrect in seven out of 14 cases.[2] For clinical symptoms, their location is much more important than their size because the risks of compressive symptoms of bronchogenic cysts in the tracheal bifurcation and paratracheal space are 68% and 57%, respectively. When the cyst is located below the hilum, only 16% of patients are symptomatic.[2] Dysphagia directly related to its compression was quite rare in the literature (Table 1). Symptomatic cysts and cysts with suspicious malignant changes, although rare, are the two main indications for surgery. An asymptomatic cyst is not an indication. For symptomatic cysts, complete resection is usually curative. Incomplete resection of the cyst may lead to recurrence.[3] For acute compressive symptoms, needle aspiration may be an alternative to temporary management. Traditionally, thoracotomy is the safest way to remove a mediastinal broncohogenic cyst because, at times, the relationship between the cyst and mediastinal structures is not clear. Other alternative strategies have been reported, including endoscopic mucosal resection.[4] With the development and maturation of thoracoscopic techniques, thoracoscopic resection provides a less invasive and still safe method to completely resect such a cyst.[5,6] We believe that thoracoscopic techniques can be safely applied for complete resection of a mediastinal bronchogenic cyst provided that the patient can understand the risks and limitations. After complete resection, long-term follow-up is still necessary for such patients.

Table 1
Dysphagia caused by bronchogenic cyst and the treatment since 1995

Author	*Year*	*Age (years)*	*Gender*	*Location*	*Symptoms*	*Treatment*
Javanthi V	1997	Adult	M	Middle oesophagus	Gastroesophageal reflux and dysphagia	Thoracotomy/ excision
Briganti V	2003	3	F	Middle oesophagus	Dysphagia	Thoracotomy/ excision
Lee SY	2008	1.5	M	Middle oesophagus	Stridor and dysphagia	Thoracotomy/ excision
Monagahn TM	2010	24	F	Middle oesophagus	Dysphagia and chest pain	Thoracotomy/ excision
Tiwari MK	2010	35	F	Middle oesophagus	Dysphagia and back pain	Thoracotomy/ excision

References

1 Coselli MP, de Ipolyi P, Bloss RS, Diaz RF, Fitzgerald JB. Bronchogenic cysts above and below the diaphragm: report of eight cases. *Ann Thorac Surg* 1987;**44**:491–4

2 Ribet ME, Copin MC, Gosselin B. Bronchogenic cysts of the mediastinum. *J Thorac Cardiovasc Surg* 1995;**109**:1003–10

3 Miller DC, Walter JP, Guthaner DF, Mark JB. Recurrent mediastinal bronchogenic cyst. Cause of bronchial obstruction and compression of superior vena cava and pulmonary artery. *Chest* 1978;**74**:218–20

4 Sashiyama H, Miyazaki S, Okazaki Y, *et al*. Esophageal bronchogenic cyst successfully excised by endoscopic mucosal resection. *Gastrointest Endosc* 2002;**56**:141–5

5 Sasajima K, Koizumi K, Onda M, *et al*. Thoracoscopic excision with mini-thoracotomy for a bronchogenic cyst of the esophagus. *Hepatogastroenterology* 1999;**46**:1785–7

6 Watson DI, Britten-Jones R. Thoracoscopic excision of bronchogenic cyst of the esophagus. *Surg Endosc* 1995;**9**:824–5