Infected Bronchogenic Cyst Treated With Drainage Followed by Resection

Yo Kawaguchi, MD, Jun Hanaoka, MD, Shoji Asakura, MD, and Takuya Fujita, MD

Division of General Thoracic Surgery, Kohka Public Hospital, and Division of General Thoracic Surgery, Department of Surgery, Shiga University of Medical Science, Shiga, Japan

Bronchogenic cysts originate from anomalous development of the ventral foregut. Although treatment of asymptomatic bronchogenic cysts remains controversial, symptomatic bronchogenic cysts should be surgically removed. We report a case of a 62-year-old man with an infected bronchogenic cyst. We drained the cyst using transesophageal endoscopic ultrasonography to control the inflammation and decrease the size of the cyst; we subsequently resected the cyst. Five months after resection, the patient was well, and computed tomography showed no evidence of cyst recurrence.

(Ann Thorac Surg 2014;98:332–4)

FEATURE ARTICLES

Although treatment of asymptomatic bronchogenic cysts remains controversial, symptomatic bronchogenic cysts should be surgically removed. However, surgical excision can be hazardous when cysts are infected because of dense adhesions to surrounding vital organs due to severe inflammation. Drainage of bronchogenic cysts is useful as a nonsurgical treatment for acute cyst decompression and reduction of inflammation. However, drainage of bronchogenic cysts alone leaves the possibility of recurrence of cyst and infection; that suggests that a combination of drainage and surgery may be useful for treating symptomatic bronchogenic cysts.

A 62-year-old man presented with a 3-day history of back pain and fever that did not respond to a 3-day course of levofloxacin prescribed by a private clinic. The chest radiograph showed an air-fluid level in the mediastinum, and chest computed tomography (CT) revealed a well-circumscribed, rounded, 90 × 70 mm diameter cyst in the subcarina corresponding with the air-fluid level and causing compression of the esophagus and both mainstem bronchi (Fig 1). We determined that the cyst had grown as a result of infection. After a 1-week course of imipenem/cilastatin provided only partial relief of symptoms, we decided to use endoscopic ultrasonography (EUS) for transesophageal drainage of the cyst and placement of a drainage tube for cyst decompression and reduction of inflammation. Gastrointestinal endoscopic examination showed extrinsic compression of the esophageal wall. The EUS examination revealed a sharply demarcated, rounded hypoechogenic lesion. Under real-time ultrasonographic imaging, a 19G needle (EZ Shot 2; Olympus EndoTherapy, Center Valley, PA) was introduced into this lesion, which was 25 cm down from the nostril. After introduction of a guidewire, a 6F catheter (Flexima ENBD; Boston Scientific, Natick, MA) was advanced approximately 15 cm into the cyst (Fig 2). A pproximately 40 mL of brown fluid was drained, and fluid cytology confirmed acute inflammation with no evidence of malignancy. Fluid culture revealed *Klebsiella pneumoniae* and *Pseudomonas aeruginosa*. The drainage tube was placed for 3 days before it was withdrawn from the nostril.

The patient's symptoms of back pain and fever disappeared soon after the drainage. Chest CT at 4 weeks after drainage showed a smaller lesion (47 × 34 mm; Fig 3); the inflammation had completely subsided, and a posterolateral thoracotomy was planned for definitive removal of the cyst. At surgery, we found that the cyst was adhered to lung, cardiac membrane, esophagus, and chest wall and required detachment from these organs. We judged that total excision of the cyst wall might be difficult, and hence decided to perform alcohol fixation of the mucosal lining to prevent recurrence from the remnant cyst wall. We introduced a contrast agent into the cyst to confirm that no fistulas with other organs existed; then we injected pure ethanol into the cyst. Eventually, we were able to remove approximately 80% of the cyst wall, although a portion of the cyst wall that was adhered to the cardiac membrane remained. Histopathology examination revealed that the cyst wall was mainly composed of fibrous tissue and showed chronic inflammatory and hemorrhagic cells, and was partly lined by stratified squamous epithelium. Immunohistochemical staining of the epithelium with CA19-9 secreted from bronchial glands was positive (Fig 4), and these findings were consistent with bronchogenic cyst. The patient was discharged 8 days after resection and has remained in

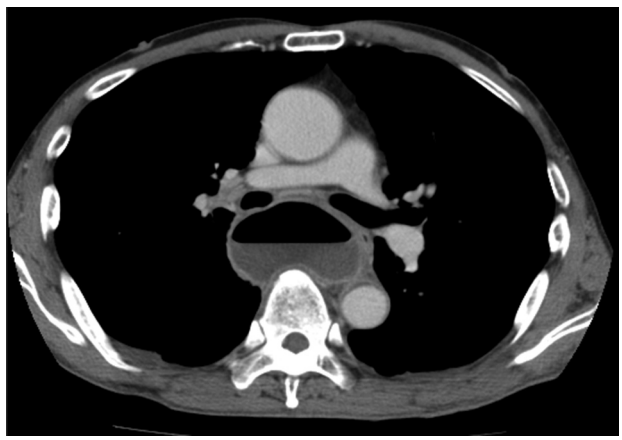

Fig 1. Chest computed tomography revealed a well-circumscribed, rounded, 90 × 70 mm diameter cyst with air-fluid level in the subcarina, causing compression of both mainstem bronchi and esophagus.

Accepted for publication Sept 17, 2013.

Address correspondence to Dr Kawaguchi, Matsuo 1256, Minakuchicho, Kohka City, Shiga, Japan; e-mail: kawaguchi1228@yahoo.co.jp.

0003-4975/$36.00
http://dx.doi.org/10.1016/j.athoracsur.2013.09.070

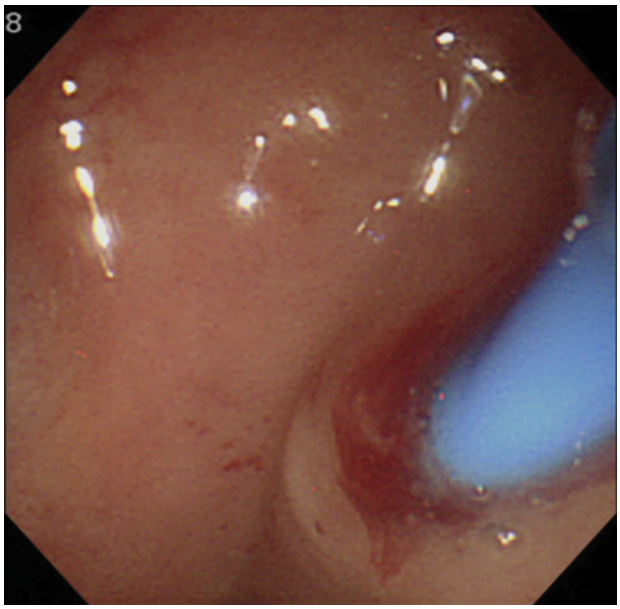

Fig 2. The cyst was drained through the esophageal wall using endoscopic ultrasonography.

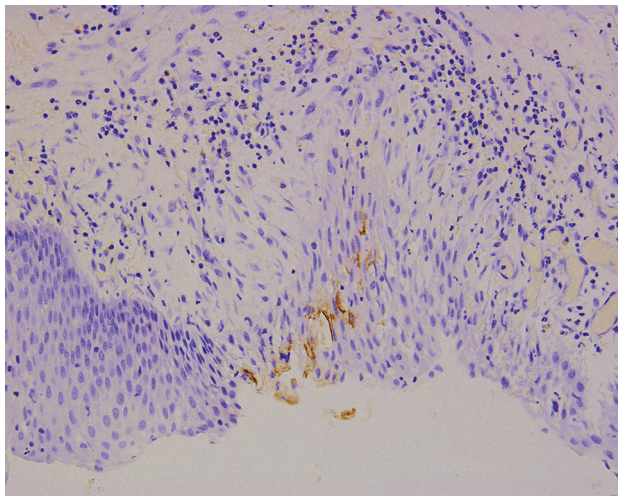

Fig 4. Histopathology examination revealed that the cyst wall consisted mainly of fibrous tissue with chronic inflammation and hemorrhage, and was partly lined by stratified squamous epithelium. Immunohistochemical staining of the epithelium with CA19-9 was positive.

stable condition for 5 months without visible recurrence of cystic lesions on chest CT scan.

Comment

Bronchogenic cysts originate from anomalous development of the ventral foregut.

Most are thought to be asymptomatic, and patients remain free of complications unless the cysts become infected or grow large enough to compress adjacent organs such as the bronchus, the heart, or the esophagus [1]. Although treatment of asymptomatic bronchogenic cysts remains controversial, most researchers agree that symptomatic bronchogenic cysts should be surgically removed.

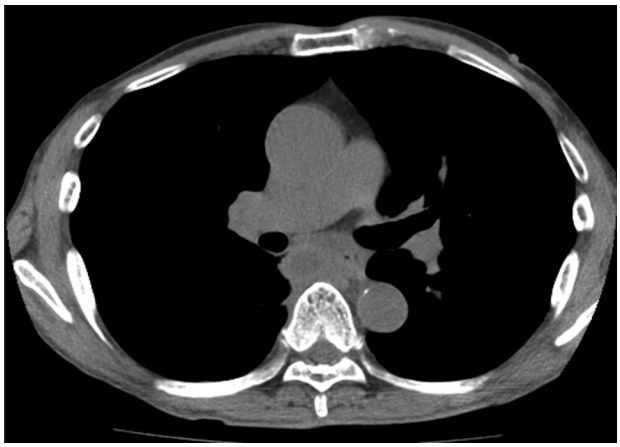

Fig 3. A chest computed tomography scan 4 weeks after the endoscopic ultrasonography drainage procedure showed a smaller lesion.

Drainage of symptomatic bronchogenic cysts has been used for acute cyst decompression and reducing inflammation in patients who are nonsurgical candidates. However, the technique is less frequently viewed as definitive treatment of symptomatic bronchogenic cysts because of the possibility of cyst recurrence. To the best of our knowledge, 10 cases of symptomatic bronchogenic cysts treated by drainage have been reported thus far. The cysts were drained by bronchoscopy, mediastinoscopy, or percutaneously under CT guidance, and in 2 cases, patients experienced recurrence, the one after 1 year [2] and the other after 7 months [3]. It is possible that the nonrecurrent cases may also carry sufficient risk of future recurrence because the reported follow-up periods were very brief, ranging from 6 months to 2 years.

No other case of drainage of a symptomatic bronchogenic cyst by EUS transesophageal drainage has been reported. The EUS transesophageal drainage allowed us to identify the structures of the mediastinum. Compared with other drainage techniques, this approach was minimally invasive, safe, easy to perform, did not require general anesthesia, and allowed us to place a drainage tube. Of the 10 previously reported cases, 1 patient had pleural effusion after bronchoscopic drainage, and this complication may have been due to a leak from the cyst [4]. In our case, we believe that dense adhesions between the cyst and the esophagus due to severe inflammation helped to prevent complications such as incomplete cyst drainage or a leak from the cyst.

The EUS-guided transesophageal drainage for cyst decompression and infection control was the first step in treating our patient. When the size of the cyst decreased, the area of the cyst wall that needed to be surgically detached from adjacent organs decreased. As a result, cyst

resection may have been easier. Moreover, we believed that controlling the infection and reducing the inflammation would contribute to lower risk of intraoperative and perioperative complications during the subsequent thoracotomy, which was performed after the cyst size was adequately reduced and the inflammation subsided. As noted, during the operation, we determined that total excision of the cyst wall might be difficult because of severe adhesions, and hence planned to perform partial resection. Masaki and colleagues [5] have reported the efficacy of ethanol injection therapy to prevent recurrence of cysts, and we performed this therapy before the partial resection. To date, there has been no evidence of recurrence from the remnant cyst wall in our patient.

Our case indicates that a combination of preoperative EUS transesophageal drainage with subsequent cyst resection may be useful for treating symptomatic and infected bronchogenic cysts.

References

1. St Georges R, Deslauriers J, Duranceau A, et al. Clinical spectrum of bronchogenic cysts of mediastinum and lung in the adult. Ann Thorac Surg 1991;52:6–13.
2. Leguen Y, Hureaux J, Gagnadoux F, Gourdier AL, Racineux JL. Treatment of a compressive bronchogenic cyst by computed tomography-guided needle aspiration. Rev Mal Respir 2005;22:481–4.
3. Sato K, Suzuki Y, Yamazaki S, Okayasu T, Hosokawa M, Fujita M. A case of bronchogenic cyst of the neck with tracheal fistula. J Jpn Surg Assoc 2010;71:1434–9.
4. Singh A, Singh S, Malpani A, Devgarha S, Singh V. Treatment of bronchogenic cyst. Surgical versus transbronchial drainage? J Bronchol Intervent Pulmonol 2011;18:359–61.
5. Masaki Y, Gomibuchi M, Haraguchi S, Hayashi K, Akiyama H, Tanaka S. Ethanol injection therapy for a bronchogenic cyst: a case report. Jpn J Chest Surg 1997;11:45–8.

FEATURE ARTICLES

Primary Pleural Malignant Melanoma With Rapid Progression

Lin Cheng, MS, Jun Nie, MD, Jinsong Li, MD, Jianjun Wang, MD, and Ke Jiang, MD

Departments of Anesthesiology, and Thoracic Surgery, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, Hubei Province, People's Republic of China

Primary pleural malignant melanoma is an exceedingly rare neoplasm. To our knowledge only 3 cases have been reported. This report presents a rare case of a primary pleural malignant melanoma with rapid progression in a 36-year-old woman. The clinical, radiologic imaging and histopathologic features are discussed here. The diagnosis of melanoma was confirmed postoperatively after the immunohistochemistry. Due to the scarcity of similar cases reported in the literature, there are no reliable data on the management and the prognosis of the disease. Therefore, histopathologic and clinical criteria are important for diagnosing primary pleural malignant melanoma.

(Ann Thorac Surg 2014;98:334–6)

Melanoma is a malignant tumor of melanocytes that are predominantly found in the skin, but can also arise in other organs and tissues of the body. However, primary pleural malignant melanoma is exceedingly rare. To our knowledge, only 3 cases have been reported [1–3]. This report presents a rare case of primary pleural malignant melanoma with rapid progression in a 36-year-old woman.

A 36-year-old nonsmoking, Chinese female presented in the emergency room with dyspnea and dizziness for 1 week. The patient denied any injury or other past medical event. Body temperature was 36.5°C, respiration rate 28/minute, heart rate 110/minute, blood pressure 90/65 mm Hg, and oxygen saturation as measured by pulse oximetry 100% on room air. On thorough and careful physical examination of the skin, no petechia, rash, or pigmentation was found. Her hemoglobin was 5.7 g/dL. The cytologic examination of pleural fluid showed many red blood cells, few lymphocytes, and neutrophils. The acid-fast bacilli test was negative on pleural fluid smear. Her erythrocyte sedimentation rate was 48 mm/hour, her c-reactive protein was 3.36 mg/L. Whole body positron emission tomography (PET) scan also showed left pleural thickening with abnormally increased multiple focal metabolism, which was suggestive of malignancy (Fig 1). No other area showed increased 18 F-fluorodeoxyglucose (^{18}F-FDG) uptake. The patient received multiple transfusions of packed red blood cells to treat anemia, after which her hemoglobin was still 6.3g/dL. Six liters of bloody pleural effusion was drained from her pleural cavity by chest tube. The patient underwent a thoracoscopic pleural biopsy under general anesthesia for diagnosis. Thoracoscopy showed firm adhesions in the left pleural cavity. There were many black oozing lamellar lesions on the parietal pleura (Fig 2). Immunohistochemical staining of the resected specimen showed extensive melan A (melanoma antigen) and S-100 proteins (Fig 3). Based on the above findings, the patient was diagnosed with primary pleural malignant melanoma. The patient declined any further treatment and died 1 month after biopsy.

Comment

Malignant melanoma predominantly arises in the skin and mucosa close to the skin; for instance, oral mucosa, intraocular mucosa, head and neck, reproductive system, rectum, and crissum. Around 160,000 new

Accepted for publication Sept 10, 2013.

Address correspondence to Dr Jiang, Department of Thoracic Surgery, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, No.1277 Jiefang Ave, Wuhan, Hubei Province, P. R. China 430022; e-mail: jiangke2013@gmail.com.

0003-4975/$36.00
http://dx.doi.org/10.1016/j.athoracsur.2013.09.065