Figure 2. Intraoperative picture of the recurrent pleural chondromatous hamartoma.

lesion seems to be a frequently used method.¹ It is assumed that once the lesion is removed it does not reoccur.^{3,5} In the presented case we have evidence to the contrary. Even though the lesion was removed with an Endopouch, there was obviously a contamination of the pleural space. This might be because of residual material of the hamartoma within the lung tissue or manipulation with the instruments. Parenchyma-sparing wedge resection of is probably the better method to resect a pulmonary hamartoma to avoid chest wall contamination.

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Large cell carcinoma arising in bronchogenic cyst

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Bronchogenic cysts are thought to originate from abnormal budding of the ventral foregut. The majority are located in the mediastinum, usually in the subcarinal or paratracheal region. Approximately 15% are reported to arise in the lung. Other locations, such as the pleura, pericardium, or diaphragm, are rare. Bronchogenic cysts are lined by

pseudostratified, columnar, ciliated (respiratory) epithelium and may contain serous fluid, mucus, blood, or purulent material. These cysts usually occur in adults but may occur in any age group. Patients usually remain asymptomatic, even though infection or bleeding produces symptoms in up to two thirds of cases. Symptoms associated with these cysts are much more common in children, in whom large cysts may compress the airways with resultant atelectasis, pneumonia, or air trapping.

We report a rare case of carcinoma arising in a bronchogenic cyst.

Clinical Summary

A 40-year-old woman with no medical history was admitted to the hospital because of radiographically proven complete pneumothorax of the right lung. Two weeks before the admission, the patient felt sudden pain in the right thorax spreading to the right shoulder with dyspnea associated with minimal physical activity. The patient's condition was growing worse with progressive dyspnea, and she felt a lack of air even during rest. She reported to the emergency department where a chest radiograph was performed, show-

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Figure 1. Computed tomographic scan of thorax: multilocular cystic lesion with a maximal diameter of 4.9 cm and a thin wall located in the center of the middle lobe of the right lung.

ing complete pneumothorax of the right lung. Arterial partial oxygen pressure before admission was 51 mm Hg. Other laboratory findings were in the normal range. On the first day of admission, thoracic drainage was performed. A control chest radiograph was performed on the same day, which showed fast and almost complete reexpansion of the lung parenchyma followed by reexpansion interstitial edema. A cavity with a thin wall approximately 4 cm in diameter was visible on the same chest x-ray film. Lung bulla was suspected, so alpha1-antitrypsin was performed, which showed normal results (1.95 g/L, range 1.2-2.2 g/L). The patient's condition was improving, so the intercostal tube was removed after 4 days. The control chest radiograph showed complete reexpansion of the lung parenchyma. A cystic lesion of the middle lobe of the right lung was still present. There was massive improvement in arterial oxygen pressure (96 mm Hg), and the patient was discharged from the hospital 5 days after admission. A computed tomographic scan was performed 5 days after discharge. The computed tomographic scan showed a multilocular cystic lesion with a maximal diameter of 4.9 cm and a thin wall located in the center of the middle lobe of the right lung (Figure 1). Surgical resection was indicated, and the patient was readmitted to the hospital 2 weeks after the discharge. Serology for echinococcus was also performed, which showed negative results. Even without the suspicion of malignancy, fiberbronchoscopy was performed, which showed the normal appearance of the bronchi. Tumor markers (CYFRA 21-1 and neuron-specific enolase) were also performed before surgery and were within the limits of those in a healthy subject.

A right intercostal thoracotomy showed a large cyst (5 cm in diameter) in the center of the middle lobe of the right lung. The cyst showed no filling. A right middle lobectomy with bronchopulmonary lymphatic dissection was undertaken. The patient left the hospital on the tenth postoperative day.

Figure 2. Cyst lined by ciliated columnar epithelium; the wall contains areas of fibroelastic tissue, smooth muscle cells, and hyaline cartilage. The cyst wall contained foci of large, atypical epithelial cells—large cell carcinoma (hematoxylin-eosin stain).

Pathologic findings showed a multilocular cyst in the middle of the right lobe with a maximal diameter of 4 cm and cavities ranging from 0.3 to 1.5 cm. Histologically, the cyst was lined by ciliated columnar epithelium, and the wall contained areas of fibroelastic tissue, smooth muscle cells, and hyaline cartilage. The cyst wall contained foci of large, atypical epithelial cells—large cell carcinoma (Figure 2). The cyst wall was intact. Bronchopulmonary lymph nodes were not affected.

Because there were no signs of dissemination of the disease, no additional therapy was applied. The patient's disease was controlled 3 months after surgery. A chest radiograph showed complete reexpansion of the remaining lung parenchyma with no signs of cystic lesion or carcinoma. Tumor markers (CYFRA 21-1 and neuron-specific enolase) were also within the limits of those in a healthy subject.

Discussion

The first bronchogenic cyst was reported by Meyer¹ in 1859. Since then, there have been numerous reports in the literature describing bronchogenic cysts. Most bronchogenic cysts originate from the mediastinum with a smaller percentage occurring in the pulmonary parenchyma. However, malignant transformation of a bronchogenic cyst is rare. The first report of malignancy associated with a bronchogenic cyst was by Moersch and Clagget² in 1947. Since then, only few well-documented cases of malignancy in intrapulmonary bronchogenic cysts have been reported in the English literature.³⁻⁶ To the best of our knowledge, this was the first case of a large cell carcinoma arising in an intrapulmonary bronchogenic cyst. Other reported cases described bronchioalveolar carcinoma, adenocarcinoma, and squamous cell carcinoma.³⁻⁶ There is not enough evidence about carcinogenesis in a bronchogenic cyst. Some evidence suggests that unstable epithelial cells in the cyst wall could have malignant potential and lead to carcinoma. Even though bronchogenic cysts are mainly asymptomatic in adults, St.

Georges and colleagues⁷ suggested that all visible bronchogenic cysts should be completely resected because the majority of them will eventually become symptomatic or complicated. As shown in our case, some bronchogenic cysts have the potential of malignant transformation.

In conclusion, we believe that complete resection of any bronchogenic cyst is justified because of the minimal risk of malignant transformation and the much greater risk of cyst-related complications (eg, pneumothorax in our case).

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A giant anterior mediastinal teratoma presenting as orthopnea and dysphagia in an adult

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A very large, mature, anterior teratoma in an adult woman presenting as orthopnea and dysphagia with bulging of the chest and neck is described.^{1,2} On excision, the mass was 27 × 20 × 11 cm in size and 14 kg in weight (inclusive of 8 L of cheesy aspirate during surgical intervention). I believe this is the largest and heaviest mediastinal teratoma ever reported in the literature.

Clinical Summary

A 20-year-old woman was referred from a remote area of a neighboring country with the diagnosis of tuberculous empyema of the left thorax and dextrocardia.³ She had received empiric antituberculous drugs for the preceding few months. She provided a history of progressive difficulty in breathing, chest pain, and fullness on the left side of the chest for the past 2 years. She also had dysphagia for solids, weight loss, and fever for the past year.

On examination, she was orthopneic, emaciated, and in tachycardia. The trachea was shifted to the right. The left side of the

chest and the left supraclavicular area were visibly prominent and bulging. No air entry or apical beat was heard on the left side of the chest. The apical impulse was in the midaxillary line on the right side of the chest.

The results of routine laboratory investigations were within normal limits. A plain chest x-ray film showed extreme displacement of the trachea, main bronchi, and heart to the right side caused by a huge mass or collection in the left hemithorax, which was seen as an opacity extending from the left supraclavicular area and pushing down the left diaphragm. Pleural aspirate was thick, purulent, and not free flowing. The cytology of the aspirated fluid showed no atypical cells, and the culture of the aspirate was sterile.

A barium swallow showed extreme displacement of the esophagus to the right and posteriorly caused by a mediastinum mass.

The fiberoptic bronchoscope revealed external compression and narrowing of all the divisions of the left main bronchus. The biopsy showed chronic inflammation, and sputum aspirate was sterile.

Ultrasonographic, computed tomographic, and magnetic resonance imaging scans of the chest showed the heart, great vessels, trachea, main bronchi, and right lung all displaced to the right side, with total collapse of the left lung caused by a large, well-defined, partially calcified mass of 25 × 20 × 9 cm in size with septum. It occupied the whole of the left side of the chest and extended from the left supraclavicular area to the left diaphragm to the right side

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