Unsuspected Lung Cancer Accompanied by Catamenial Pneumothorax

A 45-year-old nonsmoking woman with repeated coughing and dyspnea on effort was admitted to our hospital diagnosed with right-sided pneumothorax on chest X-ray. Chest computed tomography showed neither bullae nor nodules. Chest drainage failed to completely reexpand the lung, necessitating video-assisted thoracic surgery. Thoracoscopy showed pleural thickening in the apical segment without bullae or air leakage, dark-brown pigmentation of the diaphragm, and an unsuspected small nodule about 5 mm in diameter on the diaphragmatic surface of the right lower lobe. Pneumothorax was treated by mechanical abrasion of parietal pleura and upper lobe wedge resection. The lower lobe and nodule were wedge-resected using staplers. The nodule was bronchioloalveolar carcinoma of Noguchi's type B. To improve curability and check for diaphragmatic lesions, right posterolateral thoracotomy was conducted on post-video-assisted thoracic surgery day 28. Aggressive intraoperative lymph node exploration yielded no remarkable histological findings. Nonanatomical lower lobe wedge resection was done and the diaphragm with pinhole-like perforations was partially resected. The resected lung showed no cancerous tissue. Endometrial tissue was histologically confirmed in the resected diaphragm. The patient has remained asymptomatic in 14-month follow-up. This is, to our knowledge, the first lung cancer accompanied by catamenial pneumothorax.
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Key words: lung cancer, catamenial pneumothorax, diaphragmatic endometriosis

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Spontaneous pneumothorax occurs in 0.46 percent of cases of bronchial neoplasm, and that due to primary lung cancer constitutes 0.03–0.05% of the total. Lung cancer is accompanied by pneumothorax in 1.2% of all cases of spontaneous pneumothorax.[1] We report a case of unsuspected lung cancer found during thoracoscopy for catamenial pneumothorax. As far as we know from searching the literature based on Index medicus-Medline, this is the first case reported of lung cancer accompanied by catamenial pneumothorax.

Case

A 45-year-old nonsmoking woman was admitted to our hospital diagnosed with right-sided pneumothorax on chest X-ray (Fig. 1). The first symptoms suggestive of pneumothorax had appeared at the start of her menstrual periods 6 months before. Since then, she had experienced slight dyspnea on effort and a repeated dry cough. On admission, she was first diagnosed with pneumothorax. She underwent cesarean section at age 29, and subsequent occasional gynecologic examinations showed no sign of pelvic endometriosis. Chest computed tomographic (CT) scan showed neither bullae nor nodules immediately or retrospectively. Chest drainage failed to completely reexpand the lung, necessitating video-assisted thoracic surgery (VATS) showing pleural thickening of the apical segment without bullae or air leakage. The upper lobe was wedge-resected using staplers. With affected lung half-deflated, exploration of the thoracic cavity by forceps-palpation showed an unsuspected small nodule that did not rupture into the thoracic cavity on the diaphragmatic surface of the lower lobe (Fig. 2). Wedge resection of the lower lobe including the nodule was done using staplers. Endoscopy showed dark-brown pigmentation and thinning of the diaphragm near the central tendon, probably catamenial

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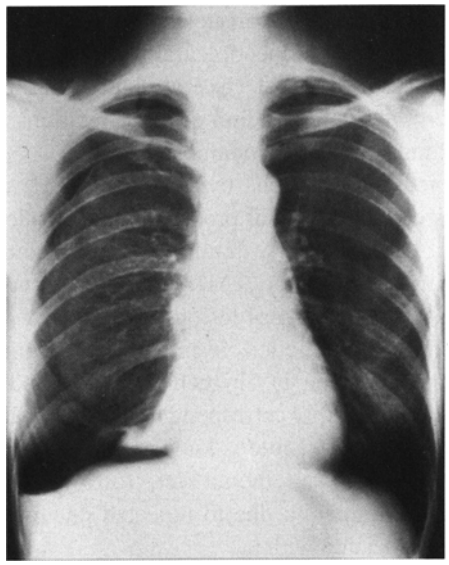

Fig. 1. Chest X-ray film demonstrating right-sided pneumothorax with pleural effusion.

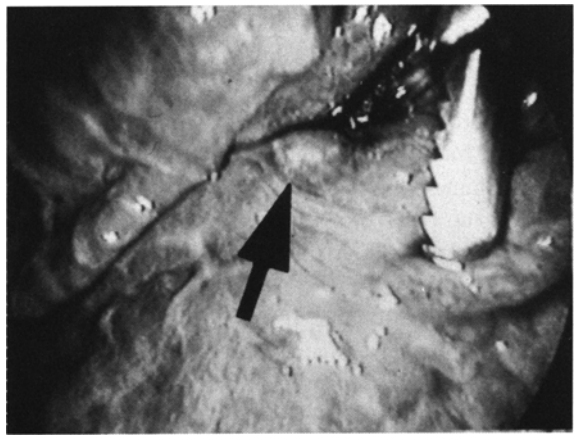

Fig. 2. Endoscopic photograph showing a small nodule (arrow) on the diaphragmatic surface of the right lower lobe. Metallic forceps used to grasp the tumor are 5 mm in diameter.

pneumothorax due to diaphragmatic endometriosis, necessitating mechanical abrasion of the parietal pleura. Histological examination of permanent sections showed the 5×3 mm nodule to be bronchioloalveolar carcinoma of Noguchi's type B (Fig. 3).[2] No specific findings were obtained from the apical specimen.

To improve curability of lung cancer and check for diaphragmatic lesions, right posterolateral thoracotomy was performed through the 6th intercostal space on post-VATS day 28. The tumor was found in the boundary between the anterior basal segment and lateral basal segment. Aggressive intraoperative lymph node

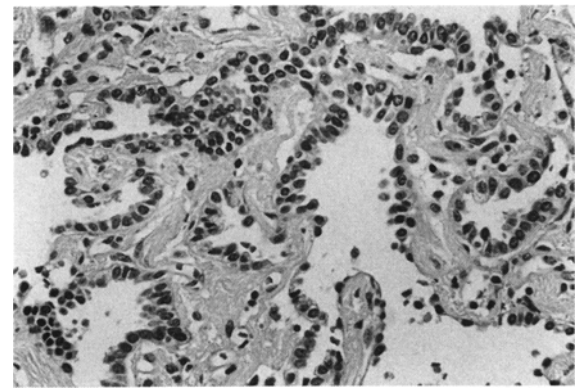

Fig. 3. Bronchioloalveolar cell carcinoma without stromal invasion or fibroblastic proliferation. H.E. stain. Original magnification: $\times$ 100.

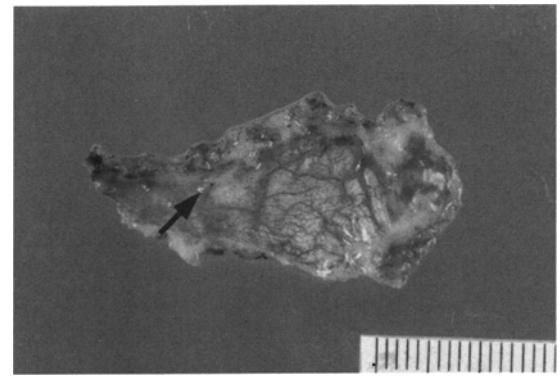

Fig. 4. Pleural surface of resected diaphragm. Note the several pinhole-like perforations (arrow).

exploration showed no remarkable findings. Additional nonanatomical wedge resection of the lower lobe was conducted with a margin exceeding 2 cm to the staplers with affected lung deflated instead of segmentectomy or lower lobectomy. The resected lung showed no remaining cancerous tissue. Thoracotomy showed several pinhole-like perforations near the central tendon of the diaphragm, which was partially resected (4×2 cm) to improve pneumothorax curability, and the defect was closed with interrupted sutures. The resected specimen showed several pinhole-like perforations (Fig. 4), and endometrial tissue was confirmed histologically (Fig. 5). These findings confirmed our diagnosis of catamenial pneumothorax. The patient has remained asymptomatic in 14-month follow-up.

Discussion

Catamenial pneumothorax is a recurrent pneumothoracic syndrome occurring mainly between 48 and 72

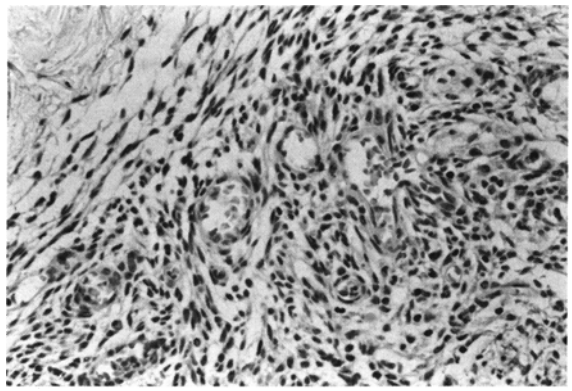

Fig. 5. Microscopic findings on resected diaphragm showing endometrial stroma extending through fibromuscular structure of diaphragm. H.E. stain. Original magnification: × 100.

hours after menstruation, first described by Maurer in 1958 and named "catamenial pneumothorax" by Lillington in 1972. This accounts for 2.8–5.6% of all episodes of spontaneous pneumothorax in women. Most women affected with catamenial pneumothorax are in the third or fourth decade of their lives, and the right hemithorax, as in our case, is affected in 90–95% of all cases.[3,4]

Four possible explanations for the pathogenesis of catamenial pneumothorax have been suggested: (1) passage of air from the genital tract via diaphragmatic congenital or endometriosis-induced defects; (2) spontaneous rupture of subpleural blebs prone to rupture during menstruation; (3) alveolar rupture due to visceral or subpleural endometrial implants; and (4) alveolar rupture due to prostaglandin-induced bronchiolar constriction. We attributed our case to endometriosis-induced diaphragmatic defects.

Surgical treatment of catamenial pneumothorax involves parietal pleurectomy, pleural abrasion, and/or partial resection of the diaphragm via conventional thoracotomy or VATS.[5] We partially resected the affected diaphragm to improve pneumothoracic curability. We use a gonadotropin-releasing hormone analog in pneumothorax recurrence, as previously recommended.[3,4]

Although it is not uncommon to encounter a patient with lung cancer associated with emphysematous lung disease, lung cancer is rarely found the first time during operation for spontaneous pneumothorax. To our knowledge, this is the first case of lung cancer reported accompanied by catamenial pneumothorax. Spontaneous pneumothorax occurring in lung cancer patients is caused by (1) rupture of dilated alveoli distal to stenotic cancer; (2) rupture of alveoli distended to compensate for atelectasis due to obstructive cancer; or (3) bronchopleural fistula secondary to rapid cancer invasion of visceral pleura.[1] In our case, however, we could not find a direct relationship between lung cancer and pneumothorax.

Atypical adenomatous hyperplasia (AAH) is now recognized as a potential precursor of lung adenocarcinoma. In some cases, minute adenocarcinomas are found in the center of AAH. In our case, no other adenocarcinoma or AAH lesions were detected in the resected lung. Allelic loss in many chromosomes has been reported even in Noguchi's type A and B tumors, which possibly correspond to bronchioloalveolar carcinoma *in situ*.[6] Little is known about the carcinogenesis of adenocarcinoma yet, for some reason, chronic inflammation due to repeated pneumothorax may be associated with it.

When lung cancer is found incidentally during surgery for other lesions, it may be difficult to change the surgical procedure, especially for such a small lesion as in our case. Indeed, limited resection for peripheral T1 N0 M0 non-small-cell lung cancer, particularly of 2 cm or less diameter, remains controversial.[7–9] Peripheral adenocarcinoma has considerable potential for lymph node metastasis, even 2 cm or less in diameter.[10] As described in Noguchi's report,[2] however, no cases of adenocarcinoma of type A or B showed lymph node metastasis. Lobectomy for such lung cancer may be avoided and limited resection may also be reasonable if there are no remarkable findings after aggressive intraoperative lymph node exploration. Histological characteristics, the anatomical location and lesion size must also be considered. In our case, the tumor, which was 5 × 3 mm in diameter and of Noguchi's type B without lymph node metastasis as far as we could tell during surgery, was located on the boundary between pulmonary segments. Taking all these into consideration, we conducted additional wedge resection with a wide margin to the tumor instead of bisegmentectomy or lobectomy, and at present, the patient remains disease-free, although intensive follow-up must be continued.

Our patient would not have been diagnosed with lung cancer before the disease advanced except for her catamenial pneumothorax. In middle-aged or elderly patients with spontaneous or catamenial pneumothorax, we should keep in mind lung cancer not detected by chest X-ray or CT, especially on the diaphragmatic surface of the lower lobe. In our case, the tumor was endoscopically difficult to detect with the fully inflated or completely deflated lung. For-

ceps-palpation definitely detected the lesion under half deflated-lung conditions, suggesting intraoperative forceps or finger palpation with the lung half deflated is useful for screening the lung for small lung cancer.

Conclusion

We report the first case in the literature we reviewed of a 45-year-old nonsmoking woman having unsuspected lung cancer with catamenial pneumothorax. The pathogenesis and treatment of catamenial pneumothorax and small adenocarcinoma of the lung remain controversial.

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