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[Case Report]

## INCIDENTAL EARLY LUNG ADENOCARCINOMA AFTER SURGERY FOR CATAMENIAL PNEUMOTHORAX

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**Abstract :** A 40-year-old female patient underwent surgery at our hospital for recurrent pneumothorax. A defect on the right diaphragm was diagnosed as ectopic endometriosis. However, air leakage continued 2 days after surgery. Chest computed tomography identified a 5-mm ground glass opacity in the right S3 field, suggestive of lung cancer. Ten days after the initial surgery, she underwent curative surgery for both pneumothorax and the lung tumor. The tumor was diagnosed as bronchioloalveolar carcinoma, but no other endometriosis was identified. The patient has remained well with no recurrence of lung cancer or pneumothorax since the second surgery.

**Key words :** lung cancer, catamenial pneumothorax, ectopic endometriosis

### INTRODUCTION

Catamenial pneumothorax is a relatively rare entity characterized by recurrent pneumothorax during menstruation. It accounts for 2.8% to 5.6% of all episodes of spontaneous pneumothorax in women<sup>1)</sup> and occurs in the third or fourth decade of life. Asymptomatic early-stage lung cancer is also occasionally encountered, even in young or middle-aged individuals. We herein report a rare case of incidentally identified early lung cancer after video-assisted thoracic surgery (VATS) for catamenial pneumothorax.

### CASE

A 40-year-old female patient was admitted to our hospital because of dyspnea at the beginning of menstruation. Chest X-ray examination showed complete collapse of the right lung (Fig. 1A). A chest computed tomography (CT) scan after chest drainage revealed pulmonary infiltration due to re-expanding lung edema after chest drainage, but no emphysematous lesions (Fig. 2A). She had visited

our hospital 9 months earlier with similar symptoms during menstruation. Catamenial pneumothorax was suspected based on her medical history, and she underwent VATS with two access ports and one window (Fig. 3A). A porous lesion without a blueberry spot on the right diaphragm was resected using staplers and was pathologically diagnosed as ectopic endometriosis with both hematoxylin-eosin staining and immunohistochemical staining (Fig. 4A, B, and C). However, air leakage was recognized 2 days after surgery and was continuous. A chest X-ray revealed an air space in the right lower field (Fig. 1B). Another chest CT scan identified a ground glass opacity (GGO) over an area of 5 mm in the right S3 segment, leading to a diagnosis of suspected early lung cancer (Fig. 2B). It was also present on the previous chest CT scan. Ten days after the initial surgery, the patient underwent a second surgery for both the pneumothorax and the lung tumor. Under small thoracotomy at the fourth intercostal space using the same port sites previously used (Fig. 3B), partial resection of the right upper lobe including the small tumor, which was detected by finger palpation, was performed. We

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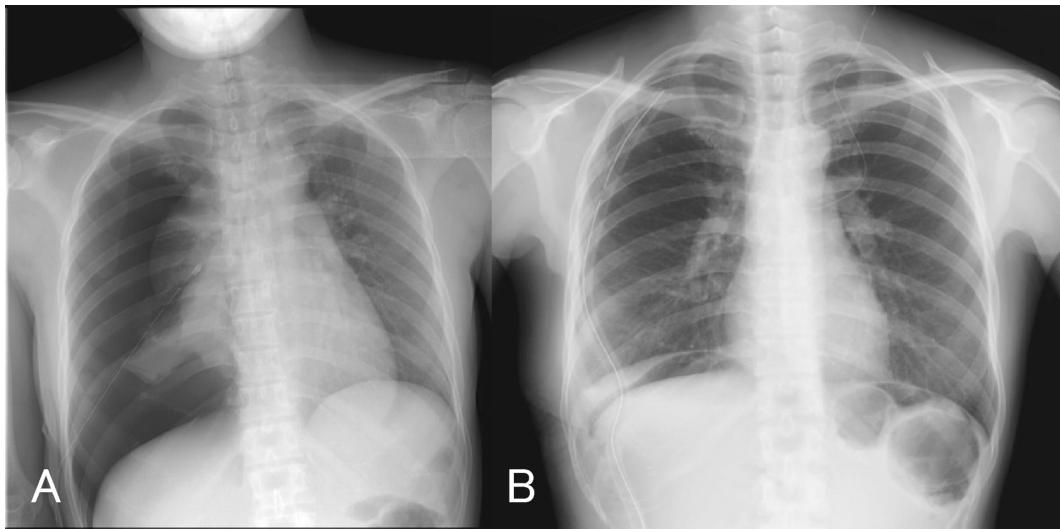


Fig. 1. (A) Chest X-ray shows complete collapse of the right lung at the time of admission. (B) Air space detected in the right lower field 2 days after first surgery.

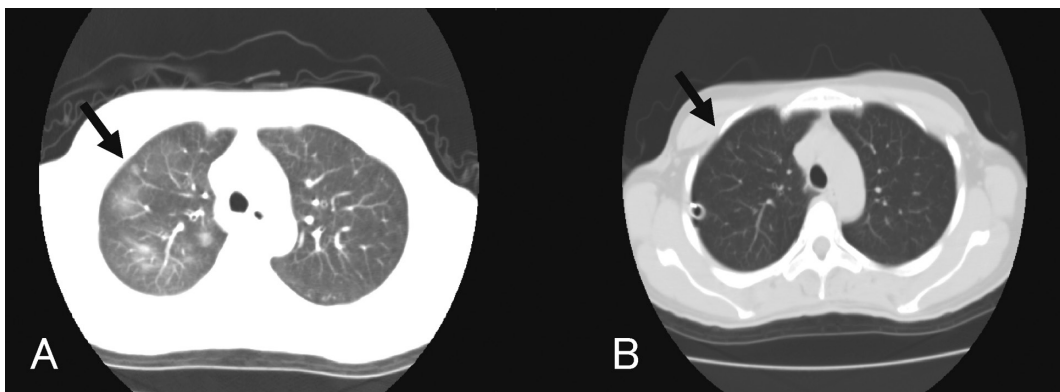


Fig. 2. (A) Chest computed tomography reveals pulmonary infiltration and no emphysematous lesions. A 5-mm ground glass opacity is detected in the right S3 field (arrow). (B) The same GGO is also detected in the same field before second surgery (arrow).

also performed partial resection of a small bulla in the middle lobe that was responsible for the air leakage. The bulla was not detected on the preoperative CT scan, and it might have also contributed to the previous pneumothorax. The tumor specimen was diagnosed during surgery as bronchioloalveolar carcinoma (BAC), and right upper lobectomy with standard node dissection of ND2a-2<sup>2)</sup> was therefore also performed on the basis of the consent form we obtained before the second surgery. The tumor was confirmed as Noguchi's type B BAC<sup>3)</sup> (Fig. 4D), but no other endometrioses were identified on the basis of pathologic findings. The final pathologic tumor staging was pT1aN0M0 stage IA<sup>2)</sup>. The patient remained well at 28 months following the second surgery, with no recurrence of lung cancer or pneumothorax and no hormonal therapy.

## DISCUSSION

We encountered a rare case of early-stage lung adenocarcinoma detected during treatment for catamenial pneumothorax. Spontaneous pneumothorax as a complication of early lung cancer is rare, comprising only 0.05% of all cases of pneumothorax<sup>4)</sup>. To the best of our knowledge, there has been only one previous report of the detection of early lung cancer with catamenial pneumothorax<sup>1)</sup>, although we presume that cases similar to ours will increase in the future with the advance of radiographic images. Spontaneous pneumothorax may occur in advanced lung cancer, and possible pathogenetic mechanisms include (a) direct tumoral invasion of the pleura, (b) rupture of a subpleural bleb in an area of obstructive emphysema, (c) an emphysematous

bullae, or (d) other unknown mechanisms<sup>5</sup>. However, direct tumor invasion or rupture of a tumor-involving subpleural bleb cannot be the cause in a case with early lung cancer. In the present case,

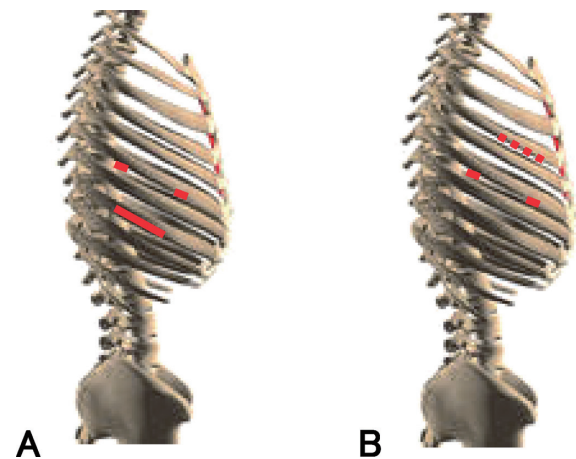


Fig. 3. (A) The initial surgery was performed using the small thoracotomy of the anterior eighth intercostal space and two port sites of the mid- and posterior axillary line at the sixth intercostal space. (B) The small thoracotomy at the fourth intercostal space was used in the second surgery (dotted line) with the previous two access ports.

lung cancer was detected incidentally after surgery for pneumothorax, and no direct relationship between the lung cancer and pneumothorax could be determined. The relationship between sex hormones and lung cancer in women was recently discussed<sup>6</sup>. While some reports have stated that the stimulation of estrogen receptor  $\beta$  (ER $\beta$ ) by estradiol leads to proliferation of lung cancer cells<sup>6</sup>, another report stated that hormone exposure is not associated with lung cancer risk<sup>7</sup>.

Catamenial pneumothorax is defined as recurrent pneumothorax that occurs during menstruation and was first described by Mauer in 1958<sup>8</sup>. The right hemithorax is affected in 90% to 95% of cases<sup>9</sup>. Its mechanisms are controversial, and hypotheses include (a) passage of intraperitoneal air through a diaphragmatic defect<sup>8</sup>, (b) alveolar rupture caused by a subpleural endometrial implant<sup>10</sup>, and (c) alveolar rupture due to bronchial spasm caused by prostaglandin F<sub>2</sub> $\alpha$  (PGF<sub>2</sub> $\alpha$ )<sup>11</sup>. Although small defects in the diaphragm and ectopic endometrial tissue were detected in the present patient, the pneumothorax originated from the rupture of a small bulla of the middle lobe. This case might thus support the following hypothesis reported by Makhija *et al.*<sup>12</sup>:

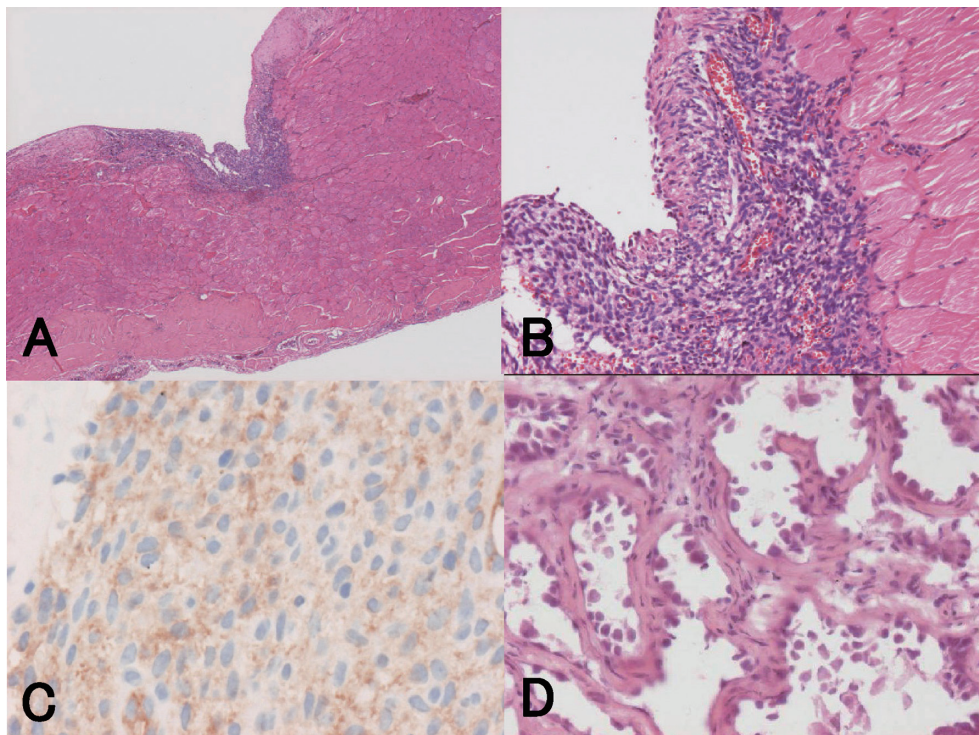


Fig. 4. (A, B) Pathologic findings of the resected diaphragm show ectopic endometrial tissue between the parietal pleura and fibromuscular structure of the diaphragm (hematoxylin-eosin;  $\times 20$  and  $\times 100$ , respectively). (C) Immunohistochemistry with CD10 reveals positive staining (anti-CD10 antibody,  $\times 200$ ). (D) The small lung tumor was diagnosed as bronchioloalveolar carcinoma, compatible with Noguchi's type B (hematoxylin-eosin,  $\times 100$ ).



endometrial cells passed through the defect of diaphragm, were implanted on the parietal or visceral pleura, and might have fallen away during menstruation. Although we could not detect endometrial cells or tissues in the resected lung tissues, we presume that the reason is that all surgeries we experienced were not performed during the menopausal period. Hormonal treatment for catamenial pneumothorax is controversial, and its long-term use is associated with adverse effects. The present patient did not give consent for the use of gonadotropin-releasing hormone analogs or oral contraceptives to prevent recurrent pneumothorax after surgery; however, hormonal therapy should be considered if a patient continues to suffer from recurrent catamenial pneumothorax.

Even in the event of pneumothorax with ectopic endometriosis in the thoracic cavity, the entire lung surface should be examined to detect bullae that might be responsible for the pneumothorax, as demonstrated by this case. In this case, the early-stage lung cancer might have been missed if the air leakage had not occurred after the initial surgery. Therefore, the whole lung should be screened intraoperatively to prevent missing small bullae, small tumors, or other lesions. The treatment for small GGO lesions remains controversial. In this case, we performed lobectomy for curative treatment with the patient's consent. However, limited surgery such as wedge resection or segmentectomy may be adequate treatment, at least for this patient. Furthermore, if the GGO in this case was detected in the contralateral lung field before or after the first surgery, it could be treated by regular monitoring rather than by curative surgery.

### CONCLUSION

We have presented a rare case of early lung cancer with ectopic endometriosis and a pulmonary bulla. In the event of pneumothorax, the whole lung should be screened, even in young and middle-aged pneumothorax patients.

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