

[ CASE REPORT ]

## A Patient with Lung Adenocarcinoma Accompanied by Whole Right Lung Torsion Induced by the Accumulation of a Large Amount of Pleural Effusion

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### Abstract:

A 68-year-old man visited a physician with a chief complaint of difficulty breathing. Right pleural effusion was noted, and he was referred to our department for a close examination and treatment. Thoracoscopy was performed under local anesthesia, and pleural dissemination of lung adenocarcinoma was noted, so a chest drain was placed. Since poor right lung inflation persisted and whole right lung torsion was observed on computed tomography, thoracoscopy-assisted thoracotomic reduction of lung torsion was performed. In this patient, the right middle lobe and anterior chest wall were adhered, suggesting that whole right lung torsion occurred when atelectasis was formed due to pleural effusion.

**Key words:** carcinomatous pleurisy, lung cancer, lung torsion, pleural effusion

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### Introduction

Lung torsion is defined as whole lung torsion or torsion of the individual lung lobes accompanied by airway obstruction and impaired blood flow using the bronchi and pulmonary blood vessels as an axis (1). It is a rare disease that occurs after lobectomy in 0.089-0.4% of cases but may also secondarily accompany trauma, atelectasis, pleural effusion, collapse, congenital defect of the pulmonary ligament, diaphragmatic hernia, and obstruction by tumor (2, 3).

We encountered a case of primary lung adenocarcinoma that secondarily caused lung torsion through carcinomatous pleurisy in a patient with no medical history of lobectomy or trauma.

### Case Report

Patient: A 68-year-old man.

Chief complaint: Difficulty breathing.

Medical history: None in particular.

Social history: Cigarette smoking, 2 cigarettes/day for 6 months.

History of present illness: The patient had been occasionally aware of shortness of breath on exertion, but he had not visited a medical institution. This time, he visited a physician for respiratory distress and chest pain, and right pleural effusion was observed on chest plain radiography. The patient was referred to our department and admitted for a close examination and treatment.

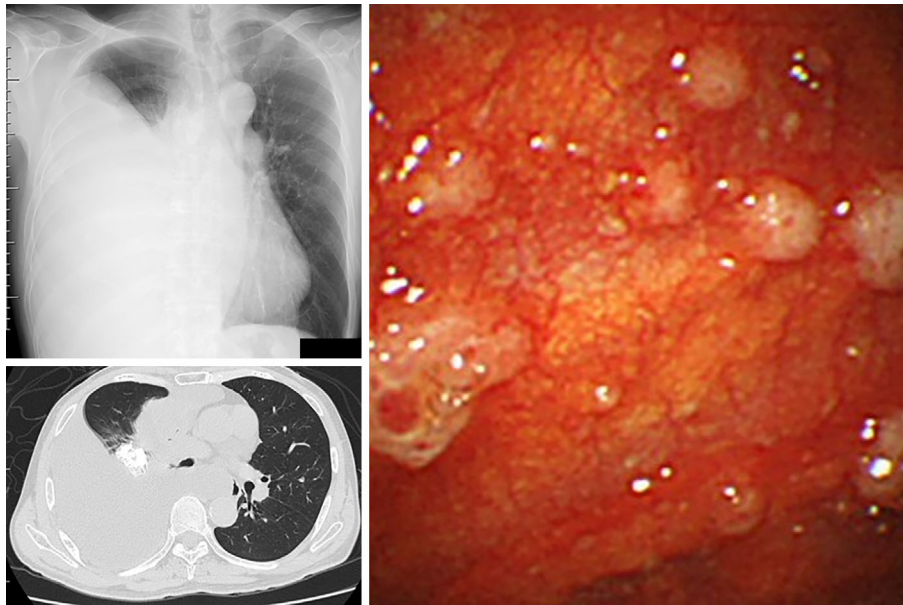
Findings on admission: Height, 165.8 cm; body weight, 54.6 kg; body temperature, 37.6°C; blood pressure, 150/114 mmHg; pulse, 120/min, regular; SpO<sub>2</sub>, 97% (under room air atmosphere); clear consciousness, decreased respiratory sound of the right lung field.

Blood test findings: White blood cell count, 7,800/ $\mu$ L; C-reactive protein, 3.74 mg/dL; Carcinoembryonic antigen, 31.6 ng/mL; Squamous cell carcinoma related antigen, 7.6

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**Figure 1.** Chest plain radiography on admission (left upper), chest CT on admission (left lower), and thoracoscopy under local anesthesia (right). A massive volume of pleural effusion was present in the right lung field, and the right lung was in a state of atelectasis due to exclusion by pleural effusion. On thoracoscopy under local anesthesia performed on the day following admission, many pleural dissemination lesions were present on the parietal pleura.



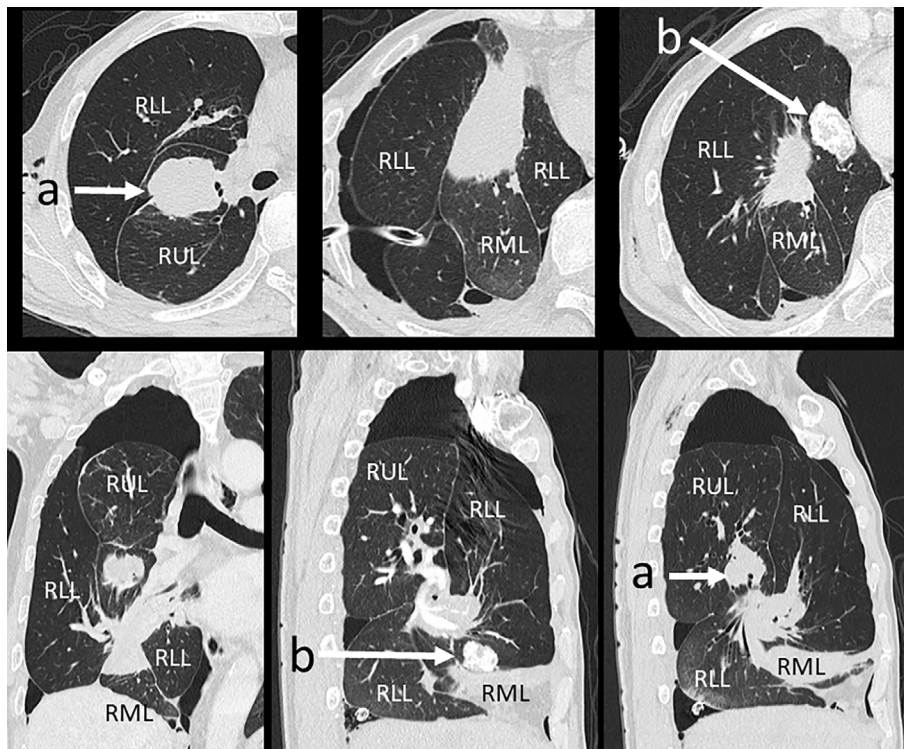
**Figure 2.** Chest plain radiography on day 7 after admission. The volume of pleural effusion had improved, but lung inflation remained poor. A mass considered the primary lesion was present in the upper lobe. Identification of the positions of the upper lobe, middle lobe, and lower lobe was difficult.

ng/mL; showing inflammatory reaction and mild elevation of the tumor marker.

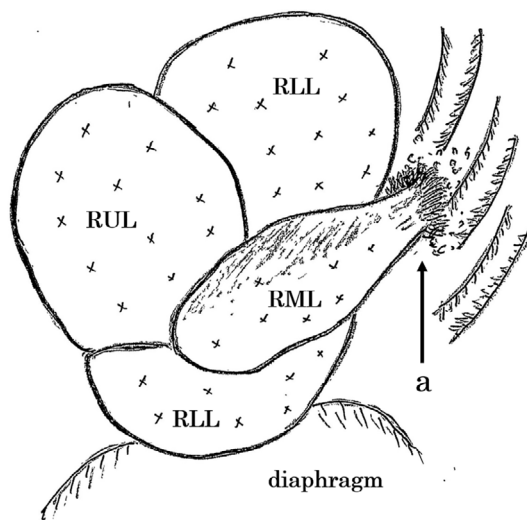
Chest plain radiography and chest computed tomography (CT) (Fig. 1): A massive volume of pleural effusion was present in the right lung field. The right lung was excluded by pleural effusion, being in a state of atelectasis.

Clinical course after admission: Thoracoscopy was performed under local anesthesia on the day following admission, and many pleural dissemination lesions were present on the parietal pleura, suggesting carcinomatous pleurisy-

induced pleural effusion (Fig. 1). After a biopsy of the pleural dissemination lesions was performed, a chest drain was placed. Chest drainage was continued, but poor lung inflation persisted on chest plain radiography (Fig. 2). On chest contrast-enhanced CT (Fig. 3) performed on day 7 after admission, the right upper lobe was positioned on the dorsal side, and a 37-mm mass was present inside. The right middle lobe was partially accompanied by atelectasis and positioned in the basal part of the lung. In addition, the right middle lobe was adhered to the anterior chest wall, and the right lower lobe partially rose on the ventral side in a state sandwiched between the middle lobe and mediastinum. The right middle lobe bronchus and right basal bronchus were partially stenosed and obstructed by torsion, and pulmonary arteriovenous and bronchial spiral torsions were observed. Based on the above findings, the patient was diagnosed with whole right lung torsion. His difficulty breathing was improved by drainage of pleural effusion, and no abnormalities were noted in the blood pressure, pulse, or oxygenation. The serum LDH level and inflammatory reaction were not elevated, and the absence of findings suggesting lung arteriovenous thrombus and pulmonary necrosis was confirmed by contrast-enhanced CT. The possibility of reperfusion injury by resolution of torsion was small, and the case was considered operable. Thoracoscopy-assisted thoracotomy reduction of lung torsion was performed on day 9 after admission. Thoracoscopic surgery was initiated, but orientation was difficult due to air trapping-induced overinflation of the lower lobe, so surgery was switched to thoracotomy. The middle lobe had adhered to the anterior chest wall and become twisted counterclockwise when observed from the



**Figure 3.** Chest CT on day 7 after admission. a: right upper lobe lung adenocarcinoma, b: right lower lobe calcified mass. On chest CT, the upper lobe accompanied by a 37-mm mass was positioned on the dorsal side, the middle lobe was partially accompanied by atelectasis and positioned in the basal part of the lung, and the whole right lung was twisted with the lower lobe rising toward the ventral side. The right middle lobe bronchus and right basal bronchus were partially stenosed and obstructed due to torsion, and spiral torsion findings of the lung arteriovenous vessels and bronchi were observed. RUL: right upper lobe, RML: right middle lobe, RLL: right lower lobe



**Figure 4.** Intraoperative findings. a: The adhered region between the right middle lobe and anterior chest wall. The middle lobe was adhered to the anterior chest wall and twisted counterclockwise when it was observed from the chest wall side, and the lower lobe twisted toward the cranial side as if it passed under the adhered region of the middle lobe. Many disseminating lesions were present on the parietal pleura. In addition, part of the middle lobe had become blackened due to atelectasis. RUL: right upper lobe, RML: right middle lobe, RLL: right lower lobe

chest wall side, and it twisted toward the cranial side as if part of the lower lobe had passed under the adhered region (Fig. 4). Although partial atelectasis was noted in the middle lobe, no pulmonary necrosis was clearly noted, and the presence of the pulmonary ligament was normal. Adhesion between the middle lobe and chest wall was dissected, torsion was resolved, and the chest was closed without fixation between the lung lobes. Many pleural disseminations were present on the chest wall, but whether or not the adhesion was due to dissemination was unclear. There were no problems with the postoperative general condition, and the resolution of torsion was confirmed by chest CT on the day following surgery. Pleurodesis was attempted on day 8 after surgery to prevent the re-accumulation of pleural effusion and lung torsion, but it was abandoned because drainage was obstructed. The drain was removed, and the patient was discharged home on day 12 after surgery. Based on the biopsy findings of pleural dissemination, the patient was diagnosed with right upper lobe ALK-positive lung adenocarcinoma (cT2bN0M1a, Stage IVA), and subsequently, alectinib treatment was initiated. The tumor size was reduced by this treatment, and as of 14 months after surgery, the course has been smooth without recurrence.

**Table. Case Reports of Spontaneous Whole Lung Torsion (Including Our Case).**

Case	Symptom	Torsion	Treatment	Cause	Outcome
51/F <sup>6)</sup>	(No description)	Whole right lung	-Torsion was resolved by exploratory thoracotomy.	-Pneumonia 10 months earlier	-Discharge on day 5 after surgery -No marked change as of 12 months after surgery
45/F <sup>7)</sup>	Dyspnea	Whole right lung	-The right middle lobe was resected by thoracotomy and torsion was resolved.	-Pseudo-Meigs' syndrome-induced of a massive volume of right pleural effusion -Right middle lobe necrosis	-No marked change as of 15 months after surgery
79/F <sup>8)</sup>	Edema/pain of the left crus	Whole right lung	-Pleural effusion was removed by thoracotomy, air was injected into the bronchus by bronchoscopy, and torsion was resolved.	-After treatment for pneumonia/pleurisy -A massive volume of pleural effusion -After thoracocentesis	-Discharge on day 6 after surgery -No marked change as of 5 months after surgery
72/M <sup>9)</sup>	Short breath	Whole left lung	-Thoracotomic excision of the whole left lung and mediastinal lymph node dissection.	-Left upper lobe mucinous adenocarcinoma -Pleural effusion	(No description)
79/F <sup>10)</sup>	Asymptomatic	Whole right lung	-Torsion was resolved by thoracoscopy-assisted thoracotomy.	-Recent medical history of pneumonia (the details are unclear)	(No description)
82/M <sup>11)</sup>	Cough Dyspnea	Whole right lung	-Thoracotomic resection of the right upper lobe	-Upper lobe central bronchial adenocarcinoma -Massive volume of pleural effusion	-Discharge on day 6 after surgery -The patient died of rapid tumor progression 2 months after initiation of chemotherapy.
76/M <sup>12)</sup>	Dyspnea Fever	Whole right lung	-Thoracotomic resection of the right upper lobe	-Giant upper lobe pulmonary adenocarcinoma -Massive volume of pleural effusion	-Discharged followed by chemotherapy
79/F <sup>4)</sup>	Dyspnea Malaise Listlessness	Whole right lung	-Video-assisted thoracoscopic resection of the right upper lobe	-Upper lobe pulmonary adenocarcinoma -Pleural effusion -Right upper lobe necrosis -After thoracocentesis	-No marked change 5 weeks after surgery
68/M (our patient)	Asymptomatic	Whole right lung	-Adhesion between the middle lobe and chest wall was dissected by thoracoscopy-assisted thoracotomy and torsion was resolved.	-Upper lobe pulmonary adenocarcinoma -Carcinomatous pleurisy-induced adhesion between the middle lobe and pleura -Massive volume of pleural effusion	-Discharge on day 12 after surgery -Initiation of chemotherapy -No marked change 14 months after surgery

F: female, M: male

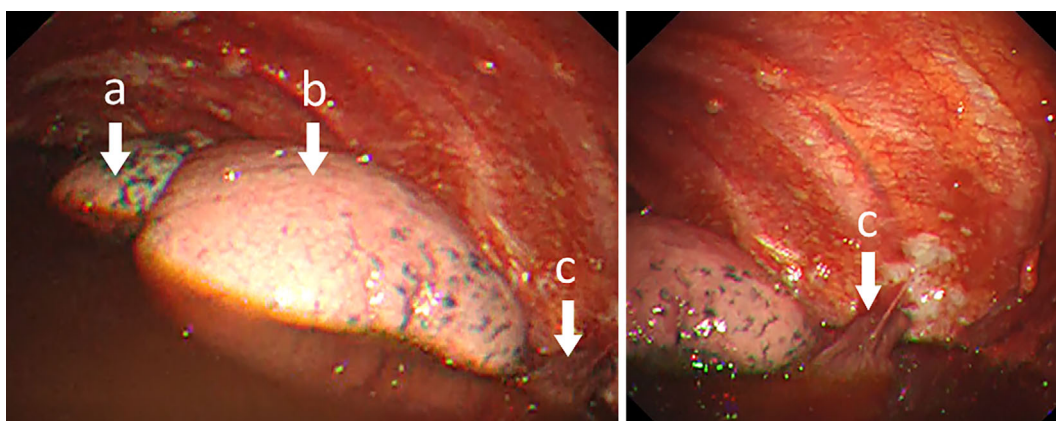
## Discussion

The cause of lung torsion is classified into three types: thoracic surgery, traumatic, and spontaneous development (2). Regarding spontaneous development, lung torsion accompanying the following events has been reported: atelectasis, pleural effusion, spontaneous pneumothorax, thoracocentesis, lung biopsy-induced pneumothorax, lung collapse, congenital defect of the pulmonary ligament, diaphragmatic hernia, obstruction by tumor, pneumonia, completely separated oblique fissure of the lung, and long bronchial pedicle (1, 2, 4, 5). The main physical findings include dyspnea, a fever, chest pain, and cough, but asymptomatic cases as in the present patient have been reported (1).

The disease is diagnosed by bronchoscopy, chest plain radiography, chest CT, and bronchography. In the present patient, poor lung inflation was observed on chest plain radiography, and abnormal positions of the lung lobes, torsional findings of the blood vessels and bronchi, and air trapping of the lower lobe were noted on chest CT. However, it may

be difficult to diagnose lung torsion by plain or contrast-enhanced CT when the lung is collapsed due to a large amount of pleural effusion, as was observed in this case. Since bronchial and lung arteriovenous torsion causes airway obstruction, venous perfusion disorder, ischemia, infarction, and necrosis and the mortality is high, rapid surgical therapeutic intervention is necessary. The best procedure to release torsion is determined based on the lung parenchymal invasiveness, and when hemorrhagic infarction and necrosis have occurred, lung resection is recommended because the risk of reperfusion injury is high (1, 4, 6, 7).

Among lung torsion cases, whole lung torsion is rare, and its prognosis is poor (1). To our knowledge, of 27 case reports in English regarding spontaneous lung torsion, 9 including our own case were spontaneous whole lung torsion (Table) (4, 6-12). Six of these cases were malignant tumor, and pleural effusion was noted in all cases. The lung lobe excluded by the massive volume of pleural effusion may have developed atelectasis, which caused lung torsion. In benign disease cases, such as cases with pneumonia, treatment was achieved through the resolution of torsion alone,



**Figure 5.** Lung lobe positions on thoracoscopy under local anesthesia. a: Right upper lobe, b: right lower lobe, c: adhered region of the middle lobe. The right middle lobe adhering to the anterior chest wall was confirmed on the diaphragmatic side of the right lower lobe.

whereas in malignant disease cases, lobectomy or pneumonectomy was performed in all cases. However, in the present case, although the disease was malignant, torsion was successfully resolved by dissection of adhesion alone without lung lobectomy, and treatment was smoothly shifted to post-treatment, being considered a valuable case.

In the present case, carcinomatous pleurisy-induced massive volume of pleural effusion excluded the lung lobe and caused atelectasis, and the right lower lobe may have risen due to a certain inducer. In addition, since the middle lobe had adhered to the anterior chest wall, air trapping of the rising lung lobe may have occurred, leading to non-resolution of torsion and whole lung torsion. Since the presence of the pulmonary ligament was normal, only the anterior right lower lobe may have risen (Fig. 4). Furthermore, atelectasis of the right middle lobe improved with dissection of adhesion of the right middle lobe and resolution of torsion, with no accompanying necrosis, suggesting that torsion had only recently formed. When images were re-confirmed, the right lower lobe was already positioned on the ventral side based on the position of the calcified lesion at S6 of the right lower lobe on chest CT on admission (Fig. 1). In addition, the middle lobe adhering to the chest wall on the mediastinal side was noted on thoracoscopy (Fig. 5). The above findings suggested that right whole lung torsion had already occurred at the time of admission.

Spontaneous lung torsion without a medical history of lobectomy or trauma is a very rare disease and may become lethal with a delayed diagnosis. Therefore, the rapid diagnosis and therapeutic intervention are important. In the present case, lung torsion had occurred at the time of admission, but due to the lack of subjective symptoms and rarity of the disease, lung torsion could not be diagnosed early by imaging alone. Spontaneous lung torsion may develop in a state with a massive volume of pleural effusion, such as carcinomatous pleurisy and pneumonia. When similar cases are observed, it

may be necessary to conduct intervention while paying attention to complications induced by lung torsion.

**The authors state that they have no Conflict of Interest (COI).**

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