

**Case  
Report**

## Adenosquamous Carcinoma of the Lung in a Patient with Complete Situs Inversus

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We present a rare case of adenosquamous carcinoma of the lung in a patient with complete situs inversus. The patient was a 76-year-old woman with the chief complaint of hemoptysis. Chest X-ray and computed tomography (CT) scans of the thorax showed a mirror image of the organs and vessels and revealed a tumor 3.5 cm in diameter, in the left lower lung field. She was referred and admitted to KKR Hokuriku Hospital, Kanazawa, Japan to undergo surgery. Bronchoscopy showed a mirror image of the usual arrangement of the bronchi, and 5 segmental branches in the left lower bronchi. During surgery, care was exercised when intubation with the Univent bronchial tube for one-lung ventilation. On thoracotomy, the gross appearance of the left lung and the arrangement of the pulmonary vessels and the bronchi corresponded to those normally found on the right side. We were successful in performing a left lower lobectomy. Postoperative diagnosis confirmed an adenosquamous carcinoma with localized pleural dissemination as p-t4n1m0, stage IIIa. Preoperative imaging, including CT, bronchoscopy, and angiographic examination of the patient, will be useful for prevention of vascular or bronchial injury during surgery in patients with complete situs inversus undergoing lung resection. Possible vascular or bronchial anomalies should always be taken into consideration while operating on these patients.

**Key words:** complete situs inversus, lung cancer, adenosquamous carcinoma, one-lung ventilation

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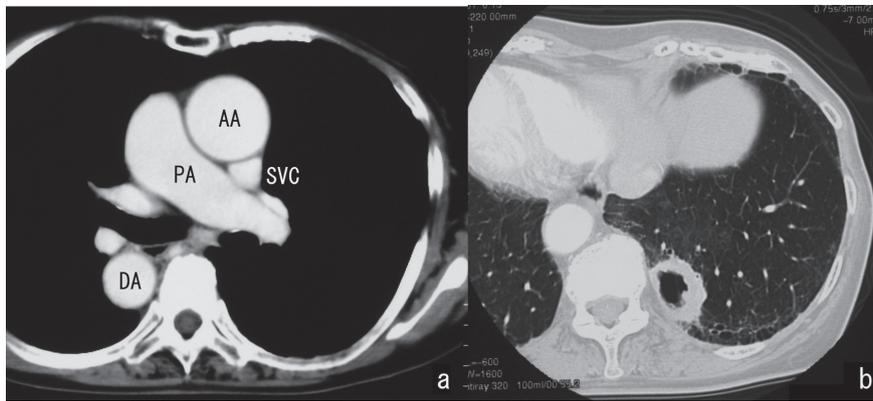
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### Introduction

Complete situs inversus is a full mirror image of the normal arrangement of the thoracic and abdominal viscera, which probably originates from an abnormal rotation of the cardiac tube during embryogenesis.<sup>1</sup> Complete situs inversus is a congenital anomaly with an autosomal recessive mode of inheritance seen in 1 or 2 people per 10,000.<sup>2</sup> There have been many reports of an association between complete situs inversus and Kartagener's syndrome,<sup>3</sup> but several reports of situs inversus in patients with lung cancer.<sup>4, 5</sup> We recently encountered a case of adenosquamous carcinoma of the lung in a patient with complete situs inversus.



**Fig. 1** **a:** Chest CT of organs and vessels in a mirror image of the usual arrangement. AA, ascending aorta; DA, descending aorta; PA, pulmonary artery; SVC, superior vena cava.  
**b:** An irregularly demarcated cavitory lesion 3.5 cm in diameter in S<sup>10</sup> of the left lung. The wall of the mass was thick and inhomogeneous, suggesting lung cancer.  
 CT, computed tomography

## Case Report

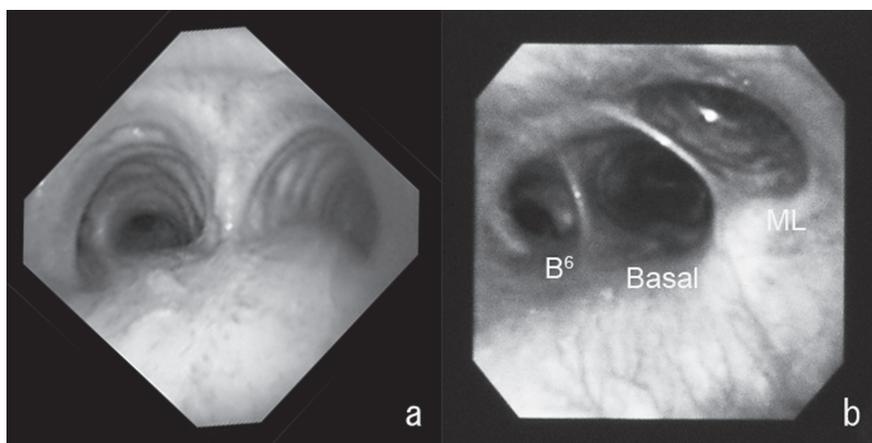
The patient was a 76-year-old woman with complete situs inversus that was identified from infancy. In September 2006, she developed bloody sputum and consulted a local clinic. Chest X-ray and computed tomography (CT) scans revealed an irregular demarcated cavitory lesion, 3.5 cm in diameter, in the posterior basal segment (S<sup>10</sup>) of the left lung; thus, she was referred to KKR Hokuriku Hospital, Kanazawa, Japan for detailed examination and treatment.

The results of hematological tests and urinalysis on admission showed no abnormalities. All tumor markers were within their normal ranges (CEA: 1.8 ng/mL; SCC: 0.6 ng/mL; CYFRA: 1.8 ng/mL; NSE: 7.7 ng/mL). Indicators of respiratory function (VC, 2.49 L; %VC, 117.5%; FEV1.0, 1.83 L; FEV1.0%, 75.0%) were within their normal ranges. The results of arterial gas analysis (PaCO<sub>2</sub>: 39.5 mmHg; PaO<sub>2</sub>: 88.0 mmHg; BE: 0.2 mEq/L; SaO<sub>2</sub>: 96.6%) were also within their normal ranges. Chest X-ray on admission revealed complete situs inversus and an irregularly demarcated mass closer to the mediastinum immediately above the left diaphragm. Chest CT scans showed a mirror image of the usual arrangement of the organs and vessels (**Fig. 1a**) and indicated an irregularly demarcated cavitory lesion, 3.5 cm in diameter, in S<sup>10</sup> of the left lung. The wall of the cavity was inhomogeneous, suggesting lung cancer (**Fig. 1b**). Findings from the nuclear medicine study revealed no sign of distant metastasis but suggested metastasis to the lymph node at the left lung hilus. Bronchoscopic examination was per-

formed and showed that tracheal and bronchial mucosa were grossly normal. The left main stem bronchus was found to be slightly veering towards the caudad-cephalad than to the right. The first lobar bronchus from the left main stem bronchus was located 2–3 cm from the carina, and the first lobar bronchus from the right main stem bronchus was located about 4–5 cm from the carina (**Fig. 2a**). The intermediate bronchus was observed on the left side (**Fig. 2b**), and 5 segmental branches were seen in the left lower bronchi. This bronchoscopy revealed no malignant findings within the visible range. In addition, malignancy was not established by brush cytology.

Based on these findings, we made a preliminary diagnosis of lung carcinoma (cT2aN1M0: stage IIA), and the patient was treated surgically in November 2006.

A tracheal tube with a mobile blocker (Univent®)<sup>6</sup> (inner diameter: 7.5 mm) was used for general anesthesia with one-lung ventilation. The chest was opened at the 6th intercostal space with a left posterolateral incision. The locations of the superior vena cava and azygos vein were mirror images of their normal distributions. The left lung was composed of 3 lobes separated by well-defined fissures (**Fig. 3**), which are typical features of the normal right side. The lowest branch of the upper pulmonary vein was the middle lobe vein, which represents the usual pattern of the right side. Bronchial anatomy also corresponded to the normal right side. A tumor 4 cm in diameter, accompanied by pleural indentation was found at S<sup>10</sup>. The tumor was wedge resected and subjected to rapid pathological examination. As the possibility of pleomorphic carcinoma was suggested pathologically, we



**Fig. 2** **a:** The first lobar bronchus from the left main stem bronchus, located 2–3 cm from the carina, and the first lobar bronchus from the right main stem bronchus, located about 4–5 cm from the carina.  
**b:** Intermediate bronchus on the left side.  
ML, middle lobe bronchus; Basal, basal bronchus



**Fig. 3** Left lung with 3 lobes, separated by well-defined fissures, assuming the formation of a right lung.

performed left lower lobectomy with lymph node dissection (ND2a). Mediastinal lymph node dissection was relatively easy because the aortic arch was absent within the left thoracic cavity. The operation took 2 h 50 min, with blood loss of 100 mL. Thus, despite the presence of situs inversus, surgery could be carried out safely through careful operative manipulations and adequate anatomical identification.

The tumor was pathologically rated as a tumor measuring  $4.8 \times 3.2 \times 2.0$  cm with an internal cavity, composed of a mixture of highly atypical poorly differentiated squamous cell carcinoma component (predominant component) and a poorly differentiated adenocarcinoma component. Minute tumor cell dissemination on the visceral pleura distant from the primary lesion was also noted. The tumor was a poorly differentiated

adenosquamous carcinoma (p-t4n1m0, stage IIIa, LN #11 metastasis (+), ly (+), v (+), dl). Lymph node metastasis was also rated pathologically as adenosquamous carcinoma.

The postoperative course was uneventful, and the patient was discharged from the hospital on the 28th postoperative day. After discharge, she received two courses of adjuvant chemotherapy with cis-platinum and vinorelbine at the referred clinic. However, she died of pleural dissemination 25 months after surgery.

## Discussion

Complete situs inversus develops due to disturbed ciliary motility during the intrauterine period.<sup>7)</sup> Twenty to twenty-five percent of cases with this anomaly present with Kartagener's syndrome, which is characterized by 3 signs: chronic sinusitis, bronchiectasis and situs inversus.<sup>3)</sup> However, the present case was free of Kartagener's syndrome. If Kartagener's syndrome is absent, complete situs inversus is often symptom-free, and ordinary surgery for such cases involves almost no problems related to management of the anesthesia.

However, when respiratory surgery, requiring one-lung ventilation, is to be performed in patients with this anomaly, special precautions are required in airway management. For example, if a double-lumen tracheal tube (DLT) designed for the left side is inserted into the left (anatomically right) bronchus of patients with situs inversus, there is a risk of obstruction of the left (anatomically right) upper lobe bronchi. Therefore, Ho et al.<sup>8)</sup> recommend the use of a tracheal tube with a mobile blocker as the first-choice when one-lung ventilation is needed in patients with situs inversus. They also proposed that if a DLT needs to be inserted, it should be bent in the opposite direction and that the tube designed for the left side should be inserted into the right (anatomically left) side and the tube designed for the right side should be inserted into the left (anatomically right) side. In the present case, a Univent<sup>®</sup> was used, allowing one-lung ventilation to be performed easily.

Local anatomy revealed that the arrangement of organs and various structures was almost a mirror image of the normal arrangement. However, some variations may be seen in the courses of blood vessels and bronchi. Therefore, for surgery in a patient with complete situs inversus, it is essential for surgeons to take extreme care in observing, identifying and handling the vascular and bronchial branches that lead to the region of planned

resection. Angiographic examination of three-dimensional CT seems to be useful for preoperative anatomical evaluation of pulmonary vessels, although we did not take such images in the present case.

As the chest was opened on the left side in the present case, mediastinal lymph node dissection was relatively easy because the aortic arch, usually present in this area, was absent. If the chest is opened on the right side in patients with this anomaly, the aortic arch, usually absent, will be present, and the recurrent nerve shows recurrence around the aortic arch, thus requiring care to avoid injury of the recurrent nerve during mediastinal lymph node dissection.

In conclusion, a review of the literature did not reveal any cases of adenosquamous carcinoma of the lung complicating complete situs inversus. Preoperative imaging, including CT, bronchoscopy, and angiographic examination of three-dimensional CT, will be useful for prevention of vascular or bronchial injury during surgery in patients with complete situs inversus undergoing lung resection. Possible vascular or bronchial anomalies should always be taken into consideration during surgery in such patients.

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