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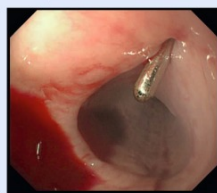
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Video-assisted thoracoscopic bisegmentectomies for double primary lung cancers in a patient with situs inversus totalis

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Abstract

Situs inversus totalis (SIT) is a rare anomaly. A limited number of reports document surgery for lung cancer in patients with SIT. We report the case of a 68-year-old man with SIT who underwent video-assisted thoracoscopic bisegmentectomies for synchronous double primary lung cancers. Preoperative evaluation of the pulmonary vessels and bronchus by three-dimensional computed tomography (CT) was unavailable owing to the patient's renal function disorder. However, the procedure was safely completed by adequate anatomic identification and careful operative manipulation based on plain CT study. His postoperative course was uneventful, and no recurrence has been observed 3 years after surgery.

Keywords Situs inversus totalis · Video-assisted thoracoscopic surgery · Segmentectomy · Three-dimensional computed tomography · Lung cancer

Introduction

Situs inversus totalis (SIT) is a rare congenital anomaly with an autosomal recessive mode of inheritance seen in 1 or 2 per 10,000 individuals [1]. It is a complete mirror image of the normal arrangement of the thoracic and abdominal viscera [2]. Vascular and bronchial anomalies are more frequent in patients with this anomaly than in the general population; therefore, surgery requires particular attention by the surgeon [3]. To the best of our knowledge, there are no reports of segmentectomies for multiple primary lung cancers in patients with complicated SIT. We report the case of a patient with synchronous double primary lung cancers with SIT who successfully underwent video-assisted thoracoscopic resections of the right apicoposterior segment and basal segment.

Case

A 68-year-old man who was diagnosed with SIT in infancy was referred to our institution for abnormal nodular shadows in the right lung field on chest radiograph during his health checkup. (Fig. 1a). He had a history of 96 pack-years of smoking. In addition, he has been taking medication for over 10 years at the family doctor, including an antiplatelet agent, for chronic cerebral infarction and diabetes mellitus with nephropathy.

The results of blood examination showed dysglycemia (hemoglobin A1c 8.4%) and renal function disorder (creatinine 2.04 mg/dl). Indicators of respiratory function suggested chronic obstructive pulmonary disorder, but were barely within normal ranges.

Computed tomography (CT) showed a complete mirror image of the normal arrangement of the organs (Fig. 1b, c). CT also indicated two nodules in the right apicoposterior segment (S1 + 2) and lateral basal segment (S9), measuring 1.5 and 2.9 cm in diameter, respectively (Fig. 1d, e), angiographic examinations of enhanced three-dimensional CT (3D-CT) were not available owing to the patient's renal function disorder. Positron emission tomography-CT revealed accumulations of fluorodeoxyglucose (FDG) in both lesions, with standard uptake values of 2.5 (S1 + 2) and 8.8 (S9). No apparent lymph nodes and/or distant metastases were shown in the routine workup.

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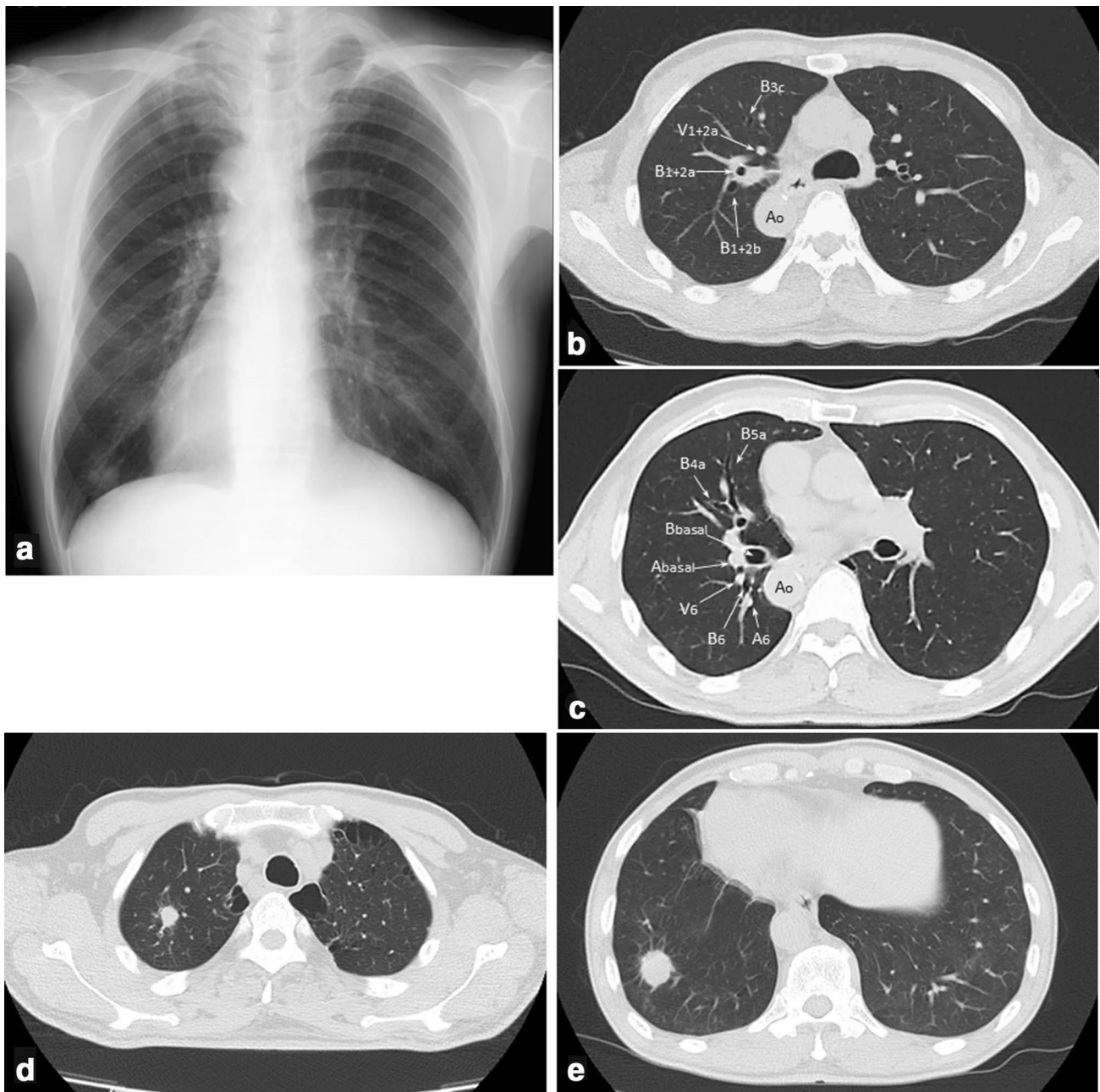


Fig. 1 **a** Chest radiograph on admission shows dextrocardia and infiltrates in the right lung field. **b** Computed tomography (CT) shows complete mirror images of the segmental veins and bronchus of the right apicoposterior segment. **c** CT shows complete mirror images of

the segmental arteries and bronchus of the right basal segment. **d** CT shows a nodule measuring 1.5 cm in diameter in the right apicoposterior segment. **e** CT shows a nodule measuring 2.9 cm in diameter in the right lateral basal segment

The histological diagnosis of squamous cell carcinoma was confirmed by transbronchial lung biopsy of the S9 tumor, while biopsy of the S1 + 2 tumor could not be obtained. Based on these findings, we made a clinical diagnosis of double primary lung cancers [c-T1b(m)N0M0, stage IA; Union for International Cancer Control, 7th edition] and planned to perform video-assisted thoracoscopic segmentectomies of the right S1 + 2, basal segment, and mediastinal

lymph node sampling. An insulin infusion sliding scale was used to control the patient's blood glucose level during the perioperative period.

One-lung ventilation was performed using a commercial double-lumen tracheal tube designed for the left side, which was inserted into the right main bronchus. With the patient in the left lateral decubitus position, the anterolateral 30-mm access port was placed in the fifth intercostal space along

the anterior axillary line, and two thoracoports were placed in the seventh intercostal space along the anterior and posterior axillary lines. The right lung was composed of two lobes separated by an incomplete interlobar fissure, as in a normal left lung. First, we performed S1 + 2 segmentectomy, dissecting in order V1 + 2, A1 + 2, B1 + 2, and the segmental boundary that was identified by the intersegmental veins (Fig. 2a). Next, we performed basal segmentectomy, dissecting in order the basal vein, bronchus, artery, and the segmental boundary due to the incomplete interlobar fissure (Fig. 2b). In addition, the subcarinal lymph node was sampled and confirmed to be cancer negative by frozen section examination. The operation took 3 h and 56 min, with blood loss of 10 ml.

The S1 + 2 tumor measuring 1.4×1.0 cm was pathologically a poorly differentiated squamous cell carcinoma, whereas the S9 tumor measuring 2.8×1.7 cm was a well-differentiated squamous cell carcinoma. Finally, based on the results of immunostaining including TTF-1, napsin A, and p63, we diagnosed his disease as double primary squamous cell carcinomas [p-T1b(m)N0M0, stage IA].

The patient was discharged on postoperative day 8 without any complications. 3 years after surgery, he is doing well with no signs of recurrence of the tumor.

Discussion

SIT develops due to disturbed ciliary motility during the intrauterine period [4]. Approximately 20–25% of patients with this anomaly present with Kartagener syndrome [5]. However, the present patient was free of Kartagener syndrome. If Kartagener syndrome is absent, SIT is often symptom-free.

In the present case, we performed video-assisted thoracoscopic bisegmentectomies with mediastinal lymph node

sampling due to the patient's extensive comorbidities. Anatomic assessment is the main issue in the management of anesthesia and pulmonary resection in patients with SIT. Recent reports advocate the efficacy of 3D-CT to evaluate the pulmonary vessels and airways [6]. However, we could not use 3D-CT in the present patient due to his renal dysfunction. When we perform lung resection including segmentectomy in normal individuals, we routinely use thin-section plain CT. In the present case, plain CT showed that the anatomy of the right pulmonary vessels and bronchus had features in common with those of the normal left side. We were able to evaluate these features in detail, as we do in normal cases, without the help of 3D-CT.

In this case, a conventional left-side double-lumen tube was inserted into the right main bronchus. In previous reports, one-lung ventilation was made possible using the opposite site of the double-lumen tube [7]. This simple method allowed us to safely perform one-lung ventilation without using a specially designed instrument for the patient.

Regarding the surgical procedure in patients with SIT, although previous reports presented cases of lobectomy or pneumonectomy, we did not find any case with segmentectomies in our literature review. As a matter of course, segmentectomy in patients with SIT tends to be more complicated than lobectomy or pneumonectomy due to the high variability of the vascular and bronchial structures and technical difficulties. Especially, it has been reported that the patient with SIT has more frequent major vessel abnormalities including cardiac malformations compared to the general population [8]. However, preoperative thin-section plain CT images showed the complete mirror image of the viscera, with no other anatomic abnormalities in the present case. Therefore, despite the presence of SIT, we judged that the procedure could be performed. In fact, we completed the bisegmentectomies according to the usual left side procedure without intraoperative complications.

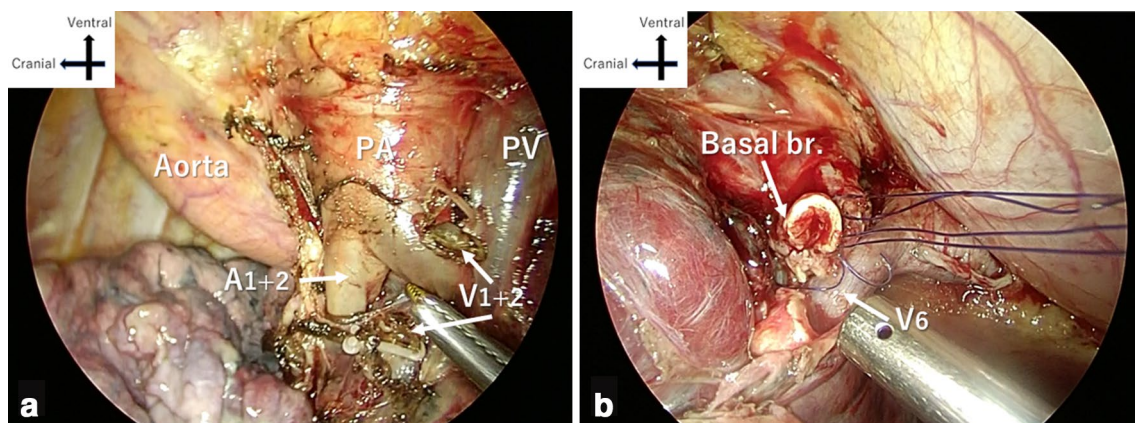


Fig. 2 Intraoperative findings. **a** The location of the aortic arch was a mirror image of the normal location. The segmental vein of the right apicoposterior segment was divided by ligation. **b** The right basal bronchus was resected using scissors and sutured with absorbable monofilaments

Conclusion

With the use of a preoperative plan based on plain CT images, video-assisted thoracoscopic segmentectomy in patients with SIT can be performed as safely as in normal individuals.

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Compliance with ethical standards

Conflict of interest The authors declare that there is no conflict of interest.

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