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CASE REPORT

# Thoracoscopic segmentectomy assisted by three-dimensional computed tomography bronchography and angiography for lung cancer in a patient living with situs inversus totalis: A case report

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**Author contributions:** All authors participated sufficiently in the work; Wu YJ is the first author; Wang YL is the corresponding author supervising the research; Wu YJ and Bao Y were the patient's thoracic surgeons and contributed to manuscript drafting; Wang YL reviewed the literature and drafted the manuscript; all authors materially participated in this work and issued final approval for this version to be submitted.

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## Abstract

### *BACKGROUND*

Situs inversus totalis (SIT) is a rare congenital condition that is characterized by a complete mirror image of the typical arrangement of the thoracic and abdominal viscera. Performing thoracoscopic segmentectomy for a patient with lung cancer and SIT is an extremely skilled and challenging surgical procedure.

### *CASE SUMMARY*

A 41-year old woman with a medical history of dextrocardia since childhood was admitted to our hospital with a mixed ground-glass opacity (mGGO) in her left lung field, discovered by computed tomography during her health checkup. In order to facilitate surgical orientation, three-dimensional computed tomography bronchography and angiography (3D-CTBA) was preoperatively carried out. The result of 3D-CTBA was consistent with the diagnosis of SIT and an mGGO in the posterior segment of the left upper lobe ($LS^2$). Surgery was conducted in accordance with preoperative 3D-CTBA and designed surgical procedure, combined with intraoperative navigation. Final pathological examination revealed *in situ* adenocarcinoma. The patient's postoperative condition was uneventful and no complications were observed.

### *CONCLUSION*

We present the first case of lung cancer in a patient with SIT who successfully underwent thoracoscopic segmentectomy assisted by 3D-CTBA. This is a new technique that covers precise confirmation and dissection of targeted structures and intersegmental demarcation, and can help achieve a meticulous anatomical

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segmentectomy.

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**Core tip:** Situs inversus totalis (SIT) is a rare congenital condition that is characterized by a complete transposition of major visceral organs. Surgical techniques in SIT patients are challenging as a result of their mirrored anatomy. A thorough understanding of segmental anatomy is thus important to avoid intraoperative accidental injuries. The three-dimensional computed tomography bronchography and angiography combined with image reconstruction with OsiriX software was found to be useful for image training before and during the surgery.

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## INTRODUCTION

Situs inversus totalis (SIT) is a rare congenital condition that occurs in an estimated 1 in 10000 people[1]. This abnormality affects all major structures within the thorax and abdomen, where major visceral organs are completely mirrored to their opposite anatomic positions. In addition, anatomical variations are more frequent than in the general population. SIT with lung cancer has rarely been reported. Herein, we present the rare case of a patient with SIT who successfully underwent thoracoscopic segmentectomy for lung cancer. Thoracoscopic segmentectomy for patients who are complicated by SIT requires particular attention from the surgeon, heralding a sizeable surgical challenge and the need for more sophisticated operations.

There are still technical pitfalls that must be overcome to achieve a meticulous and safe segmentectomy. Different imaging technology and construction software have been developed as powerful tools for the surgeons to determine anatomical structure[2-4]. Computed tomography angiography (CTA) has been established as a non-invasive, high-quality imaging tool for the evaluation of vascular structure[2]. The drawbacks of CTA in the application in the lungs are lack of stereoscopic vision, complexity of bronchial reconstruction, and exposure to ionizing radiation[2-3]. To facilitate surgical orientation and improve manipulating accuracy, we made use of three-dimensional computed tomography bronchography and angiography (3D-CTBA) to analyze anatomical variations, detect the exact location of the lung nodule, and ensure sufficient surgical margin[5-7]. To the best of our knowledge, this is the first report of thoracoscopic segmentectomy assisted by 3D-CTBA for lung cancer in a patient with SIT.

## CASE PRESENTATION

### *Chief complaints*

A 41-year-old woman was hospitalized for a mixed ground-glass opacity (mGGO) in the left lung field.

### *History of present illness*

The woman was admitted to our hospital on October 31, 2018 with an mGGO found by computed tomography during her routine health checkup (Figure 1). She had no recent history of fever, muscle ache, cough, chest pain, hoarseness, dyspnea, or blood sputum.

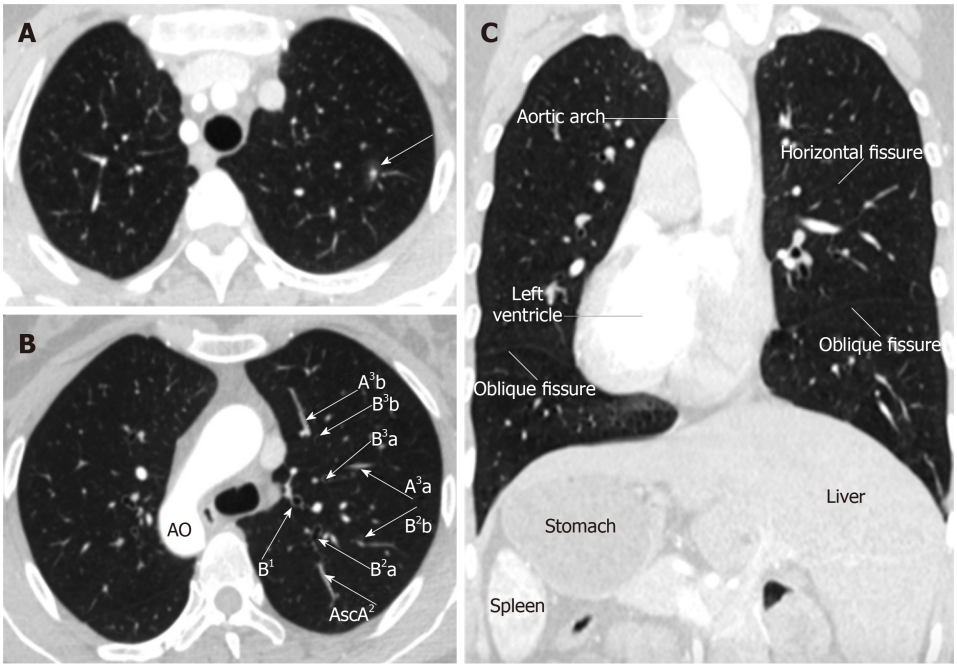


**Figure 1 Computed tomography images.** A: Mixed ground-glass opacity measuring 1.2 cm in diameter with a solid component of approximately 0.6 cm in the posterior segment of the left upper lobe ($LS^2$); B: Complete mirror image of the segmental vessels and bronchus of $LS^2$; C: Situs inversus totalis (dextrocardia, aortic arch, spleen, and liver). AscA: Ascending artery.

### *History of past illness*

The patient had, since childhood, a previous medical history of dextrocardia. However, she was free of Kartagener syndrome.

### *Personal and family history*

The patient had no history of smoking or drug abuse. Her mother died of lung adenocarcinoma while her father died of gastric cancer.

### *Physical examination upon admission and laboratory examinations*

On admission, her body temperature was 36.3 °C , pulse rate was 74 beats/min, respiration rate was 18 breaths/min, and blood pressure was 130/80 mmHg. Laboratory findings showed the serum tumor markers were within normal limits. The patient's physical examination and results of blood examination revealed no abnormal findings.

### *Imaging examinations*

Contrast-enhanced CT of the chest and abdomen was performed using a multidetector CT (MDCT) unit (Somatom Definition Flash, Siemens Healthcare, Forchheim, Germany), which not only demonstrated an mGGO in the posterior segment of the left upper lobe ($LS^2$) with no apparent lymph node and/or distant metastases, but also suggested a complete mirror image of the typical arrangement of the visceral organs (Figure 1). The mGGO, measuring 1.2 cm in diameter, consisted of a solid component, approximately 0.6 cm. The mGGO was located using a preoperative CT-guided hookwire (Figure 2).

### *Three-dimensional image reconstruction with OsiriX software*

MDCT images were recorded as digital imaging and communications in medicine (DICOM) data on a server. DICOM data were obtained and saved at a slice thickness of 1.0 mm before being transmitted to a computer and reconstructed with OsiriX software installed on a Macintosh platform (Apple, Cupertino, Calif), which acted as a multi-functional workstation for preoperative planning and intraoperative navigation. The procedure for 3D-CTBA reconstruction of the targeted structure involved using a volume-rendering method, according to the CT values of air and the definition of the thoracic surgeons. The mGGO, bronchi, pulmonary arteries, and veins were separated from each other and marked out in different colors. The result of 3D-CTBA was consistent with the diagnosis of SIT and an mGGO in the $LS^2$. We further analyzed the structure of the targeted segment, ascertained the location of the mGGO lesion, ensured the surgical margin, and finally gained a full understanding of the regional complexity (Figure 3).

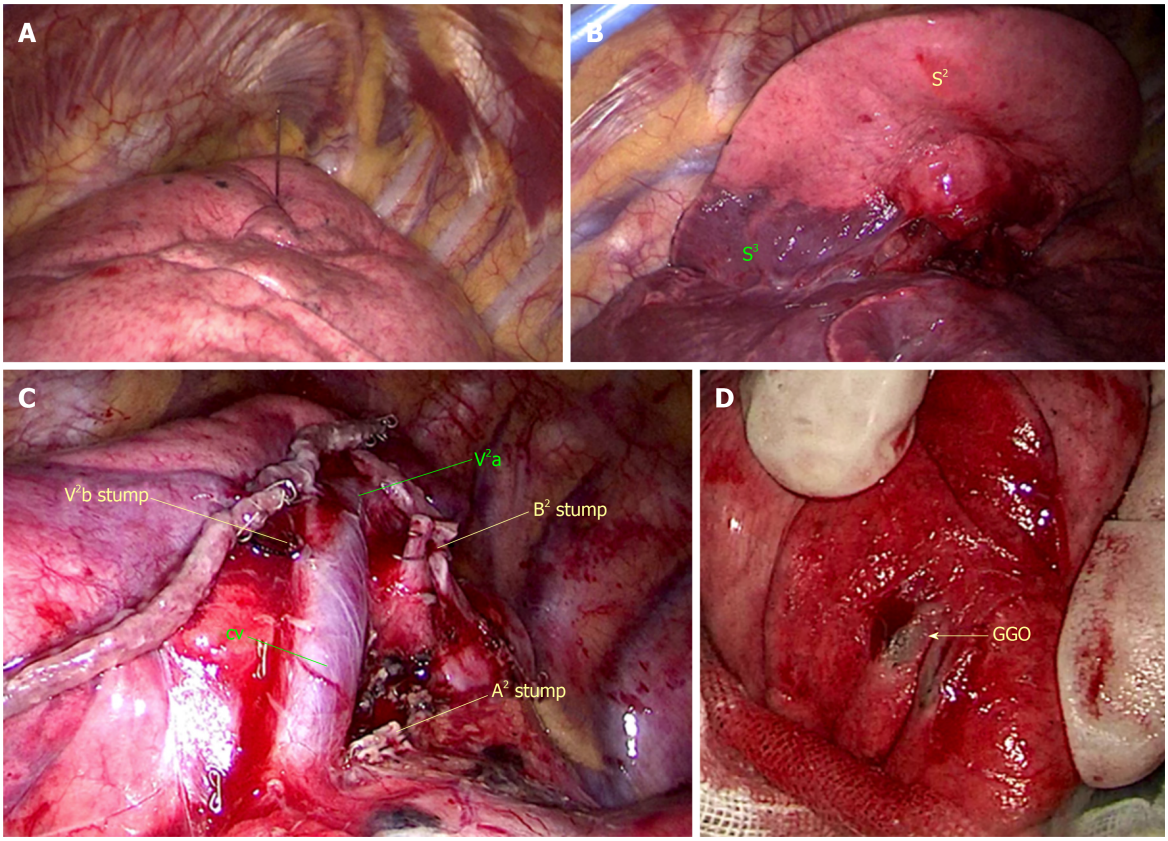


**Figure 2 Intraoperative views.** A: Computed tomography-guided hookwire localization for the mixed ground-glass opacity (mGGO) lesion; B: Intersegmental demarcation identified by an improved inflation-deflation method; C: Stumps of targeted bronchi and vessels; D: The mGGO lesion.

## FINAL DIAGNOSIS

The final diagnosis of the presented case was primary lung cancer and SIT.

## TREATMENT

Surgery was conducted according to the preoperative 3D-CTBA and designed surgical procedure, combined with intraoperative navigation. Fast-frozen pathology indicated *in situ* adenocarcinoma. Based on findings from the 3D-CTBA, we performed a thoracoscopic segmentectomy of the $LS^2$ (Figure 2). The mediastinal pleura was incised above the posterior hilum of the lung while the bronchus root of the left upper lobe was exposed. We dissected the central part of the oblique fissure to expose the interlobar artery, posterior ascending branch artery ($A^2$), and central vein (CV). In sequence, we then cut off $A^2$ and the posterior segmental bronchus ($B^2$) with a stapler. Afterwards, we separated along the CV to the distal end, exposed branches $V^2a$, $V^2b$, and $V^2c$, and dissociated the internal vein of the posterior segment ($V^2b$) by ligation cutting. Finally, we defined the intersectional boundaries between $S^1$, $S^3$, and $S^2$ using an improved inflation-deflation method (Figure 2B) and then removed $LS^2$. The Nos. 11, 12, and 13 lymph nodes were sampled. Since lymph nodes were negative for pathology, a complete mediastinal lymph node dissection was omitted. The operation took one hour and 36 min, with blood loss of 50 mL. Based on results of immunostaining, including TTF-1, napsin A, p40, and p63, final pathological examination revealed *in situ* adenocarcinoma (Figure 4). Only regular follow-up was required after surgery; the patient did not need chemotherapy or radiotherapy.

## OUTCOME AND FOLLOW-UP

The final pathologic stage was pTisN0M0 at stage 0 (Union for International Cancer Control, 8th edition). There were no intraoperative or postoperative complications such as air leak, pneumonia, or atelectasis, and the patient was discharged from hospital on postoperative day 5. No recurrence or mortality was observed during the

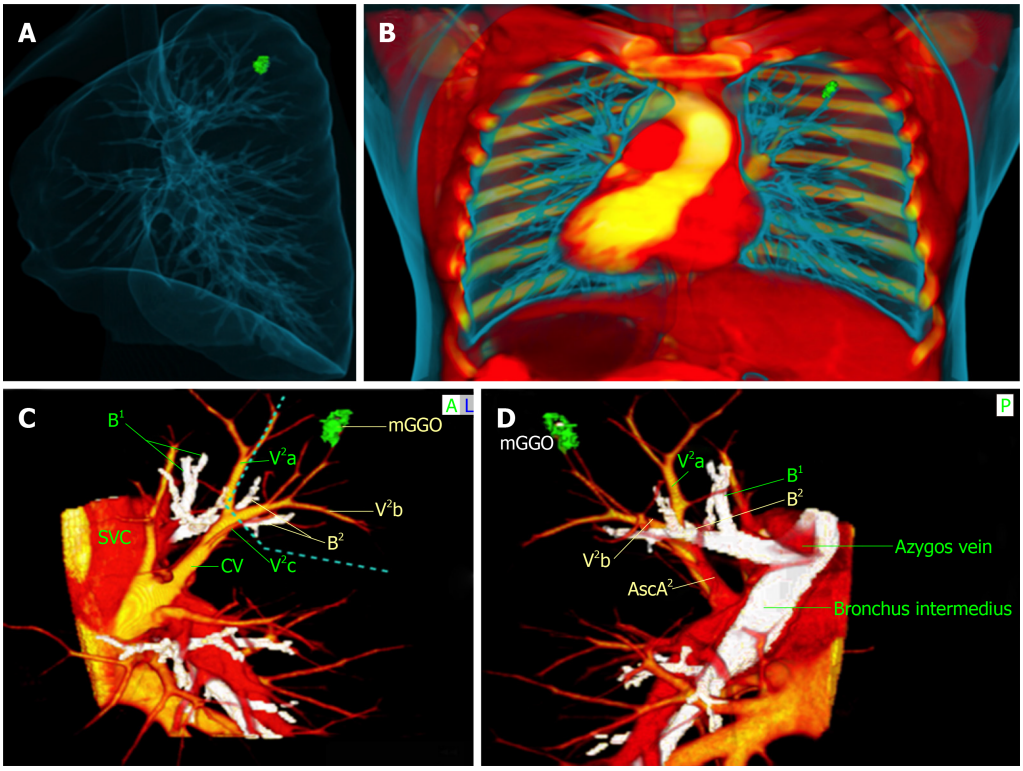


**Figure 3 Three-dimensional images reconstructed with OsiriX software.** A and B: Three-dimensional structure and spatial relation of bronchi and blood vessels; C and D: Exact three-dimensional computed tomography bronchography and angiography relationships between the mixed ground-glass opacity (mGGO) lesion and pulmonary anatomical structures. Green: The mGGO lesion; Yellow: Pulmonary veins; Red: Pulmonary artery; White: Bronchus. The blue dotted line denotes the safety margin. 3D-CTBA: Three-dimensional computed tomography bronchography and angiography; mGGO: Mixed ground-glass opacity.

follow-up period of 6 months.

## DISCUSSION

The increased popularity of health checkups as well as advances in imaging techniques have resulted in a rise of early detection rate of lung cancer and have elicited multiple questions concerning appropriate treatment for these patients[8-10]. Thoracoscopic segmentectomy for early lung cancer can maximally retain healthy lung tissue, which is beneficial for protecting postoperative lung function, improving quality of life, and reducing complications[9-10]. Since the anatomical structure of the segment is relatively complex, it is difficult to pinpoint pulmonary nodules, detect anatomical variations, identify surgical margins, and protect intersegmental veins[11-12]. As a result, it is necessary to perform thoracoscopic segmentectomy assisted by 3D-CTBA[13]. In this case, the position of the mGGO was in the left $S^2$, close to the segmental blood vessels. Careful identification in accordance with 3D-CTBA was used to clarify the location of the GGO lesion as well as its relationship to the surrounding blood vessels and bronchi, and so avoid any accidental damage.

Notably, the thoracoscopic segmentectomy of the present patient brought more challenges to surgeon owing to the transposition of the thoracic and abdominal organs to the opposite side of the body, known as SIT[1-2]. When compared to typical individuals, the anatomical structure in patients with SIT tends to be more complex and prone to anatomical variation, meaning that the risk and difficulty of surgical resection are increased. To the best of our knowledge, there are few reports of surgical resection for primary lung cancer in patients with SIT, due to its extremely low incidence[14-15].

Regarding this surgical procedure in patients with SIT and lung cancer, while previous reports have presented cases of pneumonectomy or lobectomy[16-17], we could only find two cases of thoracoscopic segmentectomy in our literature review. Wójcik *et al*[18] retrospectively reviewed 21 cases of lung cancer in patients with SIT which had been published worldwide since 1952, when the first case was described. Surgical treatment was performed through thoracotomy in ten cases and by VATS-assisted surgery only in two cases. The majority of the study group was male (20/21), and squamous cell carcinoma was the most frequent pathological type (8/21). The patient

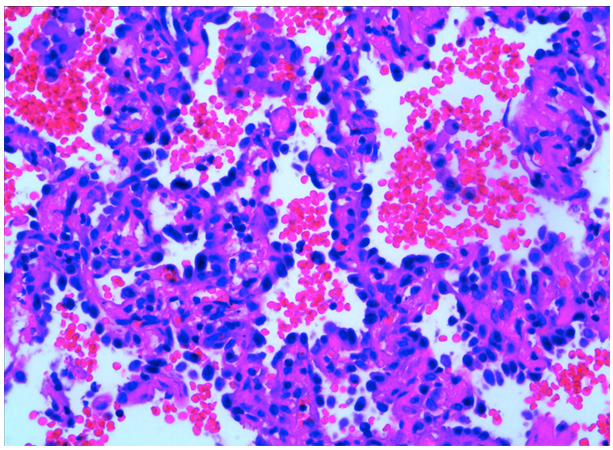

**Figure 4 Final pathology of the tumor indicated *in situ* adenocarcinoma (×200).**

in our study is female, with adenocarcinomatreated by thoracoscopic segmentectomy. Matsui *et al*[19] reported a case of thoracoscopic segmentectomy for double primary lung cancers in a patient with SIT. However, 3D-CTBA was unavailable as a result of the patient's renal dysfunction.

It is the first time we have performed thoracoscopic segmentectomy for a patient with lung cancer and SIT, with no adequate experience but a successful result. In rare cases, the selection of optimal surgical procedure as well as utilization of prior image training to simulate operation with 3D-CTBA will be beneficial to ensure surgical safety.

## CONCLUSION

Video-assisted thoracoscopic segmentectomy to treat lung cancer can be performed safely in patients living with SIT. The assistance of 3D-CTBA facilitates surgical procedures and leads to more accurate manipulation as well as precise segmentectomy for early lung cancer, which is both effective and safe.

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