

Left Uniportal VATS for Lung Decortication After Chemical Pleurodesis Under Spontaneous Breathing in Patient with Lymphangiomyomatosis – A Case Report.

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Case report

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Abstract

Background: Pneumothorax can be the first symptom of lymphangioliomyomatosis. Patients with lymphangioliomyomatosis have a higher risk of recurrence of pneumothorax. Chemical pleurodesis is a viable option to treat the recurrence, but in rare cases, it is not the solution.

Case Presentation: We present the case of a patient with lymphangioliomyomatosis undergoing a talc poudrage via video-assisted thoracoscopic surgery for pneumothorax that failed to re-expand the lung. We proposed to the patient a surgical approach to debride the lung parenchyma with the patient under deep sedation with spontaneous breathing. The patient was discharged on the 5th postoperative day. The chest computed tomography scan showed complete lung re-expansion.

Conclusion: We advocate that video-assisted thoracoscopic surgery in patients who are awake is a feasible surgical option that permits the restoration of physiological lung expansion in selected patients who underwent chemical pleurodesis and minimizes the risk of one-lung ventilation.

Background

Lymphangiomyomatosis (LAM) is a rare multisystemic disease characterized by cystic lung lesions, lymphatic abnormalities, and angiomyolipomas [1]. The spectrum of clinical presentations of LAM is variable, but often the first symptom is recurrent pneumothorax [2].

The treatment of recurrent pneumothorax in these patients is complicated by various conditions which make complete lung reexpansion difficult. The patients may exhibit chronic lung inflammation often characterized by strong pleural adhesions, and a multilocular pleural cavity. Also, the patients with LAM present significant reduction in respiratory function that does not permit safe single-lung ventilation during the operation. The purpose of paper is to show the surgical management of a young woman with LAM, respiratory failure, and recurrent left pneumothorax who was previously treated with chemical pleurodesis via video-assisted thoracoscopic surgery (VATS).

Case Presentation (Figure 1) & Surgical Technique (Video 1)

A 36-year-old woman was referred to our department with a history of progressive decline in respiratory function and a recent diagnosis of LAM. One year previously, she underwent chemical pleurodesis via VATS to treat a spontaneous left pneumothorax, but after the surgical procedure, the lung re-expansion failed. At the clinical examination, she presented with mild dyspnea during minimal exertion; a high-definition computed tomography (CT) scan showed diffuse bilateral cysts in the lung due to the LAM, and a multilocular left pleural cavity, with pleuroparenchymal adhesions and septa, and persistent incomplete re-expansion of the left lung (Fig. 2).

Surgical management was planned, taking into consideration the patient's condition and respiratory function. Cooperation with the anesthesiology team was fundamental.

The surgical procedure was performed with the patient under deep sedation with spontaneous breathing. This decision was based on 2 considerations: (1) The LAM is associated with a high risk of pneumothorax under mechanical ventilation and (2) the respiratory function of the patient did not permit safe one-lung ventilation.

Anesthesia was initiated and maintained by target-controlled infusion propofol combined with remifentanyl. High-flow oxygen was administered via a nasal mask. Radial arterial and central venous lines were placed for measuring continuous arterial blood pressure and central venous pressure.

The patient maintained spontaneous breathing throughout the operation, with stable hemodynamics and a respiratory rate of 18–20 bpm/min.

The patient was placed in a left lateral decubitus position. Thoracoscopy was performed using a single 2-cm skin incision at the 7th intercostal space near the midaxillary line. Video exploration showed a pleural cavity with multiple fibrous pleural adhesions forming a multilocular space and a visceral pleural rind that did not permit lung reexpansion. Dissection of the parietal fibrous pleural rind was performed by an ultrasound energy device (Ultracision®, Ethicon Endo-Surgery, Inc., Somerville, NJ, USA), and blunt dissection was used for visceral pleural rind decortication.

The visceral fibrous pleural rind was incised to identify the plane between the fibrous pleural rind and the lung parenchyma; then blunt separation of the 2 layers was performed. After the removal of the visceral pleural rind, the adhesions between the lung and the anterior and posterior mediastinum and the diaphragm were separated to free the lung as much as possible. This kind of dissection was performed with the combined use of an ultrasound energy device and blunt dissection.

Finally, meticulous hemostasis and aerostasis were obtained; 1 chest drain was placed posteriorly, and the complete reexpansion of the lung was monitored by video.

The chest tube was removed on the 4th postoperative day, and the patient was discharged in good respiratory condition. On a chest CT scan performed 1 month postoperatively, the left lung was completely re-expanded; at the clinical examination, the patient did not exhibit dyspnea with moderate exertion.

After surgical treatment, the patient was discharged on the 5th postoperative day. A chest CT scan done 1 month postoperatively showed complete resolution of her left-sided pneumothorax (Fig. 3).

Discussion And Conclusion

Our patient, who was a woman in her childbearing years with pulmonary manifestations and a history of spontaneous pneumothorax, presented with the classic demographic features of LAM [3]. When she was referred to our department, she presented a particular situation: After an episode of pneumothorax, she underwent chemical pleurodesis, but the procedure was not effective. The patient had an incomplete re-

expansion of the lung that was incarcerated in a visceral fibrous pleural rind caused by the pleurodesis. At this time, surgical debridement was mandatory.

To prevent the risk of one-lung ventilation, in accordance with the anesthesiologist, we chose deep sedation with spontaneous breathing [4]. For the surgeons, this technique simplified the identification of the plane between the visceral pleural rind and the lung parenchyma.

Decortication and debridement of the lung permitted normal pulmonary re-expansion, which was fundamental for avoiding pulmonary infection and for maintaining the respiratory function as well as possible.

LAM may be mistaken for other pulmonary disorders due to similarities in the initial presenting symptoms, such as restrictive or obstructive lung disease and may therefore be inadequately treated [5]. Patients with this disease are at a greater risk of developing recurrent spontaneous pneumothorax due to smooth muscle proliferation, airway narrowing, and alveolar damage. Spontaneous pneumothorax is estimated to occur in 10% of patients who have cystic lung disease. Patients with LAM who experience spontaneous pneumothorax have a higher risk of recurrence. Pleurodesis is a viable treatment option [6] but if the surgical procedure fails, reintervention for pleural decortication and lung debridement can be considered.

In conclusion, we advocate that, in these cases, awake VATS performed by a skilled thoracic surgeon is a feasible surgical option.

Abbreviations

LAM – Lymphangiomyomatosis

VATS - Video-Assisted Thoracoscopic Surgery

CT - Computed Tomography

Declarations

Ethics approval and consent to participate

Not applicable. This case report describe the use of a routinary procedure in a particular clinical setting with the aim of show the management of critical complication.

Consent for publication

The authors confirm that informed consent for publication was obtain from the patient and it is available on request.

Availability of data and materials

The authors declare that the data supporting the findings of this study are available within the article and its supplementary information files.

Competing Interests

The authors declare that they have no competing interests

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None

Author's Contributions

MS and JV were the major contributors in writing the manuscript. MB and EM performed surgical video clip. MA and FV planned the study design.

All authors read and approved the final manuscript.

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Figures

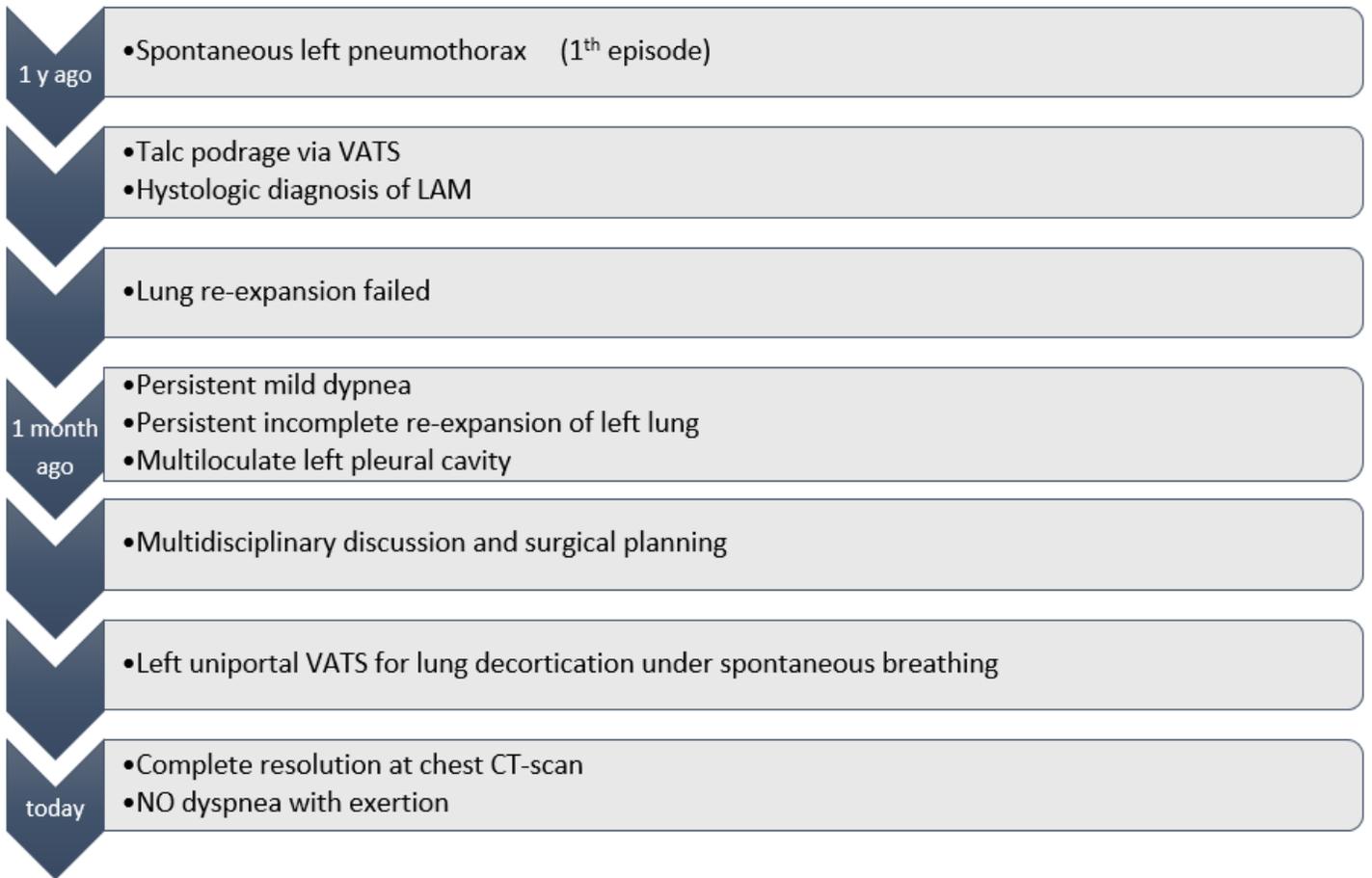


Figure 1

Timeline

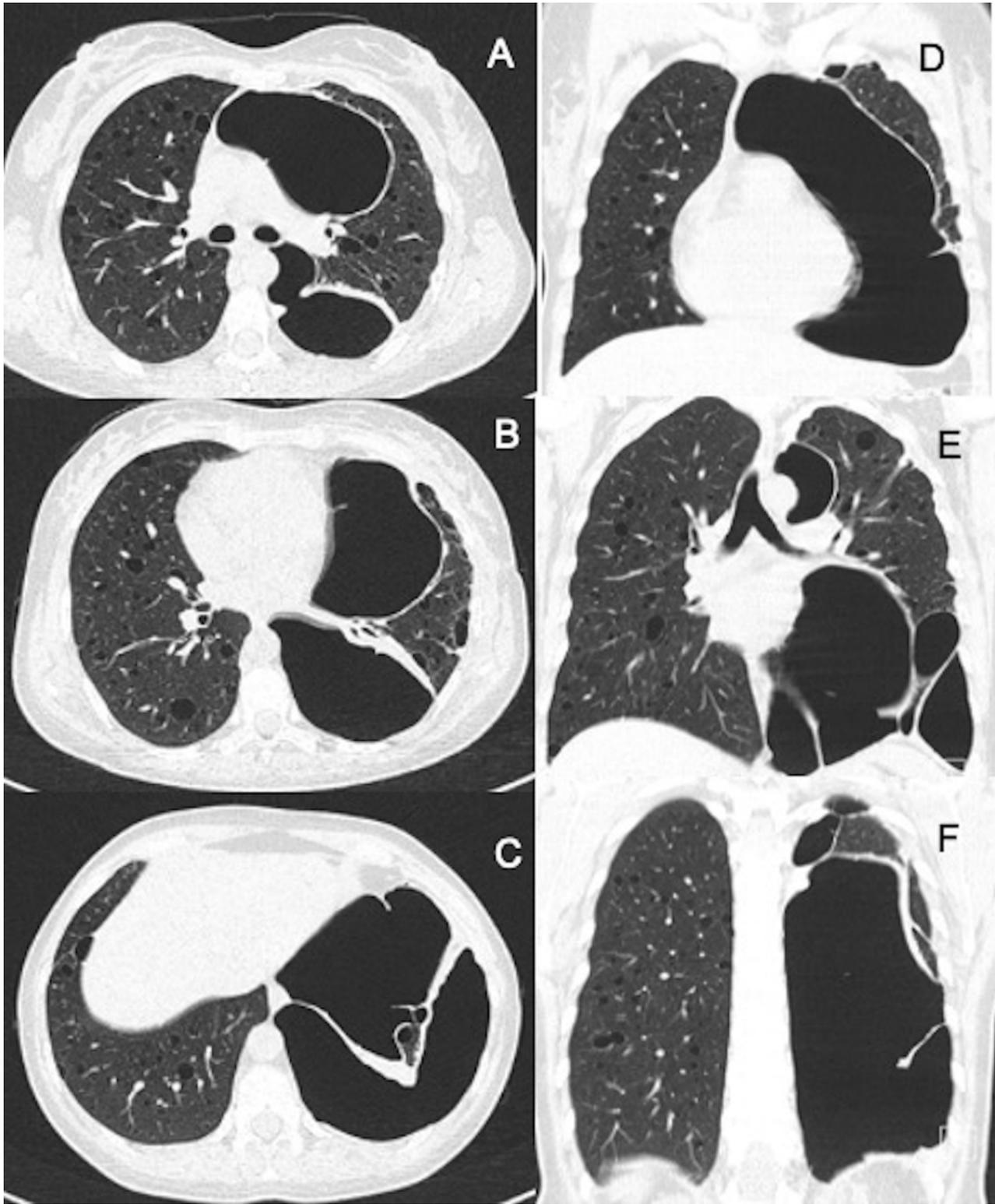


Figure 2

Preoperative CT-scan in axial view (A, B, C) and coronal view (D, E, F). The images show multilocular pleural cavity and pleuroparenchymal adhesions.

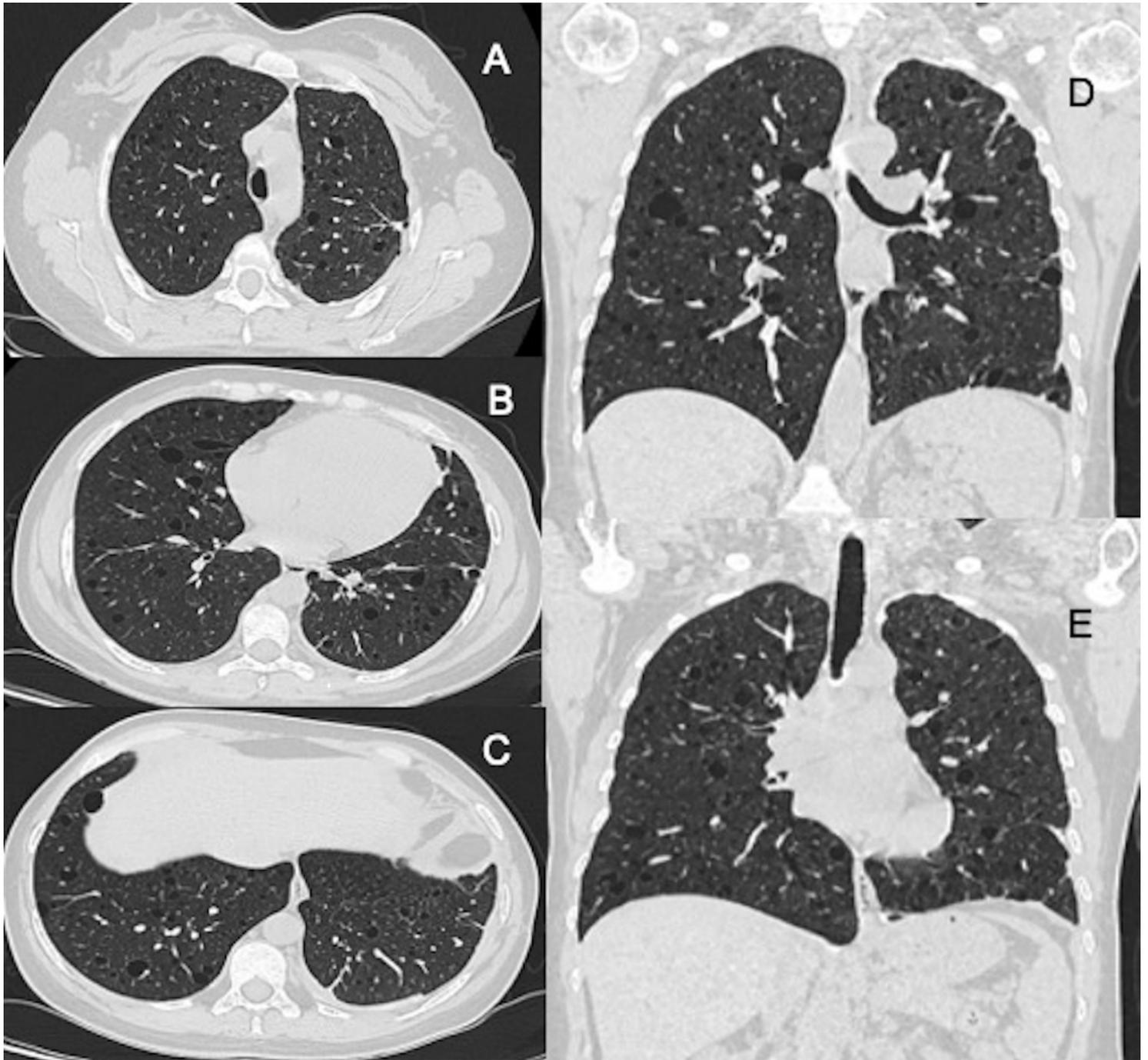


Figure 3

Postoperative images, after 1 month from surgery, show a complete expansion of the left lung and diffuse bilateral lung cysts due to the LAM (axial scan A, B, C and coronal scan D,E)

Supplementary Files

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