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Case Report

A Total Pleural Covering Technique in Patients with Intractable Bilateral Secondary Spontaneous Pneumothorax: Report of Five Cases

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Abstract
We herein present five cases of bilateral intractable secondary spontaneous pneumothorax associated with chronic severe lung diseases that were successfully treated with a modified form of a previously reported surgical procedure, the "total pleural covering technique," under video-assisted thoracic surgery. We performed the total pleural covering technique modified with a preceding coverage of air-leak points with polyglycolic acid sheets. In this series, the median length of surgery was 106 min (range: 67–220 min) on the unilateral side (10 sides). No significant surgical complications were observed, but one patient died on day 23 after the operation, due to respiratory insufficiency on the basis of the underlying lung disease. The remaining four patients have been followed up regularly (mean follow-up time: 23 months; range: 1–54 months) and there has been no recurrences of pneumothorax. We believe that the total pleural covering technique is a useful method; however, special attention should be paid to the underlying disease in order to identify patients who would be most likely to benefit from the procedure.

Key words Thoracoscopy · Secondary spontaneous pneumothorax · Total pleural covering technique

Introduction

Bilateral secondary spontaneous pneumothorax (SSP) occurring as a complication of underlying lung diseases is often intractable and fatal. Chemical pleurodesis is one of the most frequently employed therapeutic procedures for intractable SSP. However, pleural symphysis is still associated with a high recurrence rate in patients with SSP, due to several lung diseases including lymphangioleiomyomatosis (LAM).[1] Moreover, lethal complications, including acute respiratory distress syndrome, have been reported in association with talc poudrage.[2] In recent years, the total pleural covering (TPC) technique, which refers to a surgical procedure that covers the entire visceral pleura using regenerative oxidized cellulose mesh under video-assisted thoracoscopic surgery (VATS), has emerged as an effective treatment for pneumothorax secondary to LAM.[3] Kurihara et al. have reported that the mechanism by which TPC can be used to prevent recurrence of the pneumothorax is associated with thickening of the visceral pleural surface, and that despite this thickening, no restrictive impairment of lung function was experienced.[3] The aim of the present study was to describe our experiences with a modified form of TPC in five patients (10 lateral sides) with simultaneous or nonsimultaneous bilateral SSP.

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Case Reports

Patients

The clinical features of these five patients are summarized in Table 1.

Case 1
A 17-year-old male patient, with a history of two occurrences of left pneumothorax and four occurrences of right pneumothorax, was referred to our hospital complaining of back pain. He had undergone bullectomy under VATS four times at another hospital, and histological examination of the resected specimens had revealed a diagnosis of pulmonary eosinophilic granuloma. Bilateral TPC was performed.

Case 2
A 21-year-old woman complaining of back pain consulted a local hospital. A chest X-ray showed bilateral

Table 1. Clinical features of the five patients with secondary spontaneous pneumothorax treated with a modified total pleural covering technique

		Case				
		1	2	3	4	5
Age (years)		17	21	34	27	25
Sex		M	F	M	F	F
Preoperative therapy		2 op	1 op	–	1 op 2 pleuro	2 op 2 pleuro
Underlying lung disease		EG	LAM	BHD	LAM	BO
Pneumothorax	R	2	2	1	5	6
	L	4	4	1	2	2

op, operation; pleuro, chemical pleurodesis; EG, eosinophilic granuloma; LAM, lymphangioleiomyomatosis; BHD, Birt-Hogg-Dubé syndrome; BO, bronchiolitis obliterans; R, right; L, left

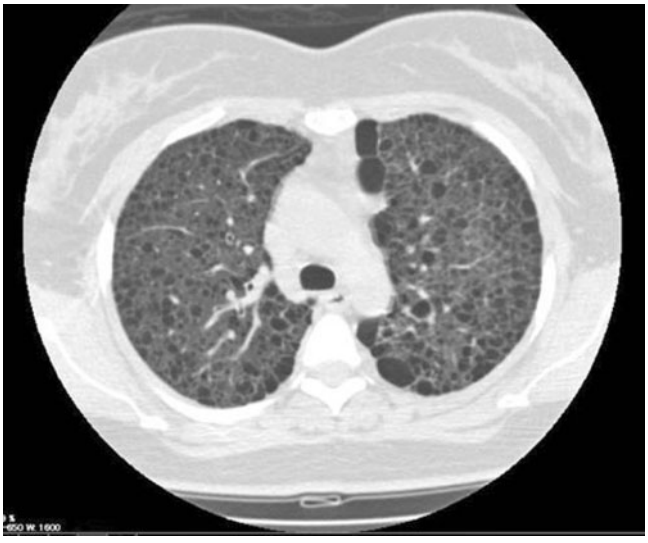

Fig. 1. Case 2. Computed tomography scan showing diffuse cystic changes in bilateral lung fields, compatible with lymphangioleiomyomatosis

pneumothorax. Chest computed tomography (CT) revealed diffuse cystic changes in the bilateral lung fields, which indicated a diagnosis of LAM (Fig. 1). She was subsequently referred to our hospital and we performed TPC on the left side (Fig. 2) because of air leakage from the left drainage tube. The right-side air leakage emerged after the operation, and TPC on the right side was performed on day 13 after the first operation.

Case 3

A 34-year-old Afghan man complaining of dyspnea was referred to our department. Chest CT showed multiple thin-walled cysts in the bilateral lung fields. Because his family history included that his brothers had experienced spontaneous pneumothorax repeatedly, we suspected a diagnosis of Birt-Hogg-Dubé syndrome, and TPC on the left side was performed. Five months after the operation, he developed right pneumothorax. TPC was performed on the right side.

Case 4

A 28-year-old woman at the 9th week of gestation presented to a local hospital complaining of dyspnea and right chest pain. A chest X-ray showed pneumothorax on the right side. Chest CT revealed a number of blebs in the apices of the bilateral lung fields, and a bullectomy was performed. Pathological examination of the resected specimen established a diagnosis of LAM. The patient developed recurrent right pneumothorax on day 6 and developed a left pneumothorax on day 12 after the operation. We performed simultaneous TPC on the both sides.[4]

Case 5

A 25-year-old woman had been suffering from bronchiolitis obliterans (BO) due to graft-versus-host disease after bone marrow transplantation (BMT) for acute myelogenous leukemia. Simultaneous bilateral pneumothorax occurred. Because of continuous air leakage on the left side, a bullectomy was performed. However, the continuous air leakage was not controlled and she was referred to our hospital. Although the patient showed severe dyspnea due to restrictive ventilatory dysfunction, her left lung was not expanded because of severe air leakage and we therefore decided to perform TPC.

Surgical Technique for TPC

The indication for TPC was intractable pneumothorax after surgery, or chemical pleurodesis due to intractable chronic progressive pulmonary diseases with diffuse multiple fragile cysts. After insertion of a thoracic epidural catheter and induction of general anesthesia, a double-lumen endotracheal tube was introduced and the patient was placed in the lateral decubitus position. Under single-lung ventilation, a 5-mm port was inserted

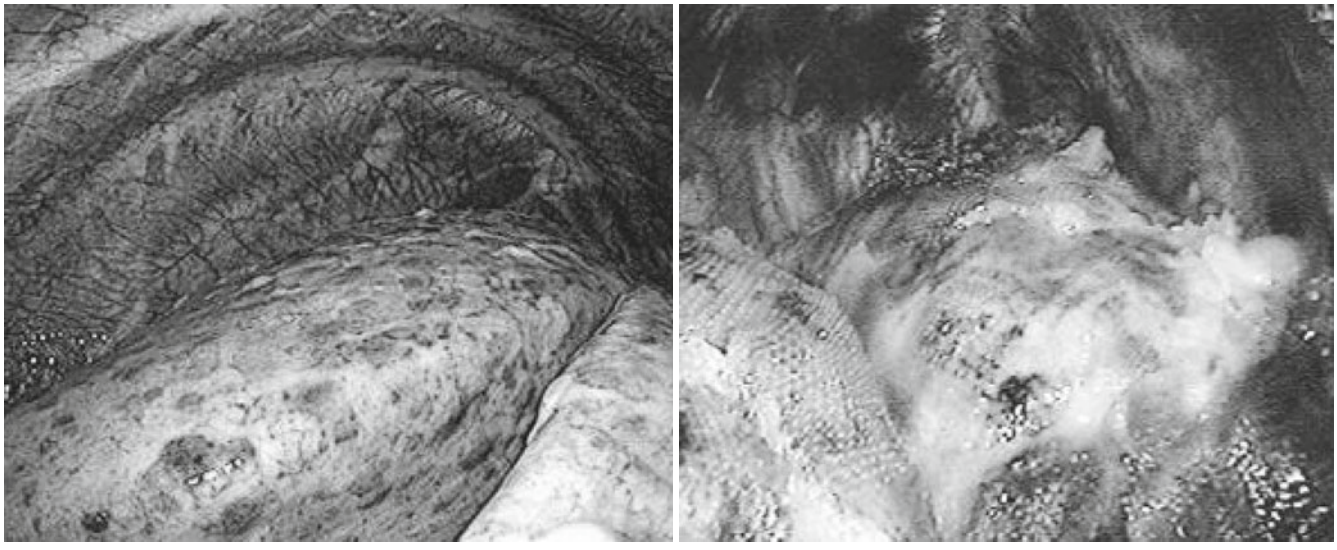

Fig. 2. Case 2. **a** Thoracoscopic findings showing multiple fragile blebs on the surface of the left lung. **b** The lung surface covered with regenerative oxidized cellulose mesh with fibrin glue

in the 7th intercostal space at the mid-axillar line. Subsequently, 11.5-mm ports were placed in the 6th intercostal space at the postero-axillar line, the 5th intercostal space at the shoulder blade underline, and the 4th intercostal space at the mid-clavicular line. The LTF thoracoscope (Olympus Optical, Tokyo, Japan) was inserted through a flexible introducer and polyglycolic acid (PGA) sheets (Neoveil; Gunze, Kyoto, Japan) were affixed to the pleural surface around air-leak points with or without stapling the area when air-leak points were detected. Total pleural covering using regenerated oxidized cellulose mesh (ROCM) (Surgicel; Johnson & Johnson, New Brunswick, NJ, USA) was applied as previously described by Kurihara et al.[3]

Once the lung deflated, we inserted the ROCM with thoracic forceps then, under the halfway-inflated lung, rolled and affixed it to the entire surface of the visceral pleura, including the interlobular and diaphragmatic surface. The lung was then fully inflated, and an additional cover was applied to the area that was uncovered after full lung inflation. We used the modified solution described by Kinoshita et al.[5] in which 10 ml each of fibrinogen solution and thrombin solution diluted with 30 ml of saline (Bolheal; Chemo-Sero-Therapeutic Research Institute, Kumamoto, Japan) were used to spray the entire ROCM surface. An atomized diluted fibrinogen solution was sprayed, and next an atomized diluted thrombin solution was applied with thoracoscopic forceps. Finally, both solutions were sprayed simultaneously onto the lung surface as a mixed aerosol using the Bolheal Spray Set (Chemo-Sero-Therapeutic Research Institute). When bilateral TPC was required for simultaneous bilateral SSP, we changed the position of the patient and applied TPC on the contralateral side in a similar manner. After the operation, the chest drainage tube was removed when the lung was fully expanded without air leakage, and drained pleural fluid decreased to less than 200 ml in a 24-h period. If air leakage continued for more than 5 days, chemical pleurodesis with 5 KE of OK-432 was performed.

Results

In the present series, the median length of surgery was 106 min (range: 67–220 min) on the unilateral side. The mean duration of the postoperative chest drainage was 9.1 days (range: 1–25 days). No significant surgical complications were observed. Postoperative air leakage prolonged for more than 5 days was seen in two patients (3 of 10 sides) and chemical pleurodesis with OK-432 was applied for both of these patients. In these three sides, the pleurodesis was successful. The air leakage in the patients was minimal and well controlled by the pleurodesis. One case (case 5) died on day 23 after the second (right side) surgery, due to respiratory insufficiency on the basis of BO after BMT. The remaining four patients have been followed up regularly (mean follow-up time: 23 months; range: 1–54 months) and there have been no recurrences of pneumothorax. The surgical results are summarized in Table 2.

Discussion

We herein presented five patients with bilateral intractable SSP who were successfully treated with the TPC

Table 2. Surgical features of the five patients with secondary spontaneous pneumothorax treated with a modified total pleural covering technique

		Case				
		1	2	3	4	5
Length of surgery (min)	R	116	135	67	110	81
	L	100	220	58	90	79
Air leakage	R	–	+	–	–	–
	L	–	+	–	+	–
Postoperative chemical pleurodesis	R	–	+	–	–	–
	L	–	+	–	+	–
Maximum CRP (mg/dl)	R	11.0	18.1	3.4	13.5	22.3
	L		10.7	9.8		22.8
Removal of the drain (days)	R	1	24	6	13	3
	L	2	25	6	6	5
Recurrence	R	–	–	–	–	–
	L	–	–	–	–	–
Prognosis		Alive	Alive	Alive	Alive	Died (POD 23)

R, right; L, left; CRP, C-reactive protein; POD, postoperative day

technique. All of our cases had severe pulmonary diseases including LAM, chronic eosinophilic granuloma, BHD, and BO. We modified the original TPC by at first covering the pleural surface around air-leak points with PGA sheets, with or without stapling the area. Polyglycolic acid sheets have been reported to be an ideal material for staple-line reinforcement,[6] and we believe that it is a more reliable material for controlling air leakage than ROCM. Next, TPC was applied using ROCM, which is less expensive than PGA sheets. Although pleurodesis was required for minimal air leakage after the TPC in two patients, bilateral pneumothorax in all patients was well controlled and no recurrence of pneumothorax has been observed. There were no intraoperative or postoperative deaths due to surgical complications in our series. One case (case 5), however, died on day 23 after the operation due to respiratory insufficiency. Although respiratory insufficiency on the basis of BO was quite severe in this case, we decided to control air leakage by surgical intervention because of lung collapse due to the fact that the severe air leakage was progressing and had not been controlled by pleurodesis. Despite successful TPC on the both sides that controlled the air leakage, the patient's hypercapnia was not recovered and she died on day 23 after the second surgery because of respiratory insufficiency. When the lung has expanded in cases of bilateral SSP, it is recommended that attempts be made to instill a sclerosant through a chest tube or bullectomy.[7] If these procedures are not available or not effective, consideration might subsequently be given to performing TPC.

In conclusion, TPC was applied safely to selected patients with simultaneous or nonsimultaneous bilateral SSP. The procedure was well tolerated and did not result in major surgical complications. We believe that TPC is a safe and reliable procedure for management of intractable SSP. However, careful attention must be paid to the underlying disease in order to determine whether patients will benefit from this procedure.

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