—Case Reports—

Video-Assisted Thoracoscopic Surgery for Catamenial Pneumothorax: A Report of Five Cases Treated by Diaphragm Reefing with and without Chemical Pleurodesis

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Background: Catamenial pneumothorax (CP), the most common manifestation of thoracic endometriosis syndrome, is a rare form of primary spontaneous pneumothorax that occurs in women of reproductive age. Although CP is usually treated surgically or with hormonal therapy, there is no standard treatment and recurrence is common. We performed video-assisted thoracoscopic surgery (VATS) for five patients with CP from 2014 to 2023.

Methods: Our surgical basic procedure (BP) was as follows. VATS was performed by using one port site for the camera and a 5-cm mini-thoracotomy. Perforated holes or blueberry spots were detected on the diaphragm, and lesions were ligated with an endoscopic loop. After reefing by ligation, the diaphragm was covered with a polyglycolic acid sheet. Three methods were used: (A) BP only (one case); (B) BP with tetracycline plus OK432 (two cases); (C) BP with tetracycline (two cases); because the use of OK432 (an anti-cancer agent) was disallowed in 2019 in our Institutional Review Board rules for patients with benign disease.

Results: Age ranged from 33 to 45 years (mean, 38.6±5.1 years), and CP occurred on the right side in all cases. Recurrence occurred in only two of the five cases (one case each for procedures B and C).

Conclusion: Our procedure (diaphragm reefing by ligation and covering with a polyglycolic acid sheet) was a more convenient and more effective treatment for CP. (J Nippon Med Sch 2025; 92: 463–467)

Key words: catamenial pneumothorax, video-assisted thoracic surgery, chemical pleurodesis, hormonal therapy

Introduction

Catamenial pneumothorax (CP), the most common manifestation of thoracic endometriosis syndrome (TES), is a rare form of primary spontaneous pneumothorax (SP) that occurs in women of reproductive age.

Surgery and/or hormonal therapy are usually used to treat CP; however, there is no established treatment, and the recurrence rate is high.

Five patients with CP underwent video-assisted thoracoscopic surgery (VATS) and reefing with covering with a polyglycolic acid (PGA) sheet of diaphragm at our hospital. The surgical procedure with and without chemical pleurodesis was evaluated.

Materials and Methods

Patients

We performed VATS for 339 cases at our hospital during a 10-year period (January 2014 to December 2023). Fifty of the 339 (13.2%) patients were female, five of whom (10% of females) were diagnosed with CP via pre-

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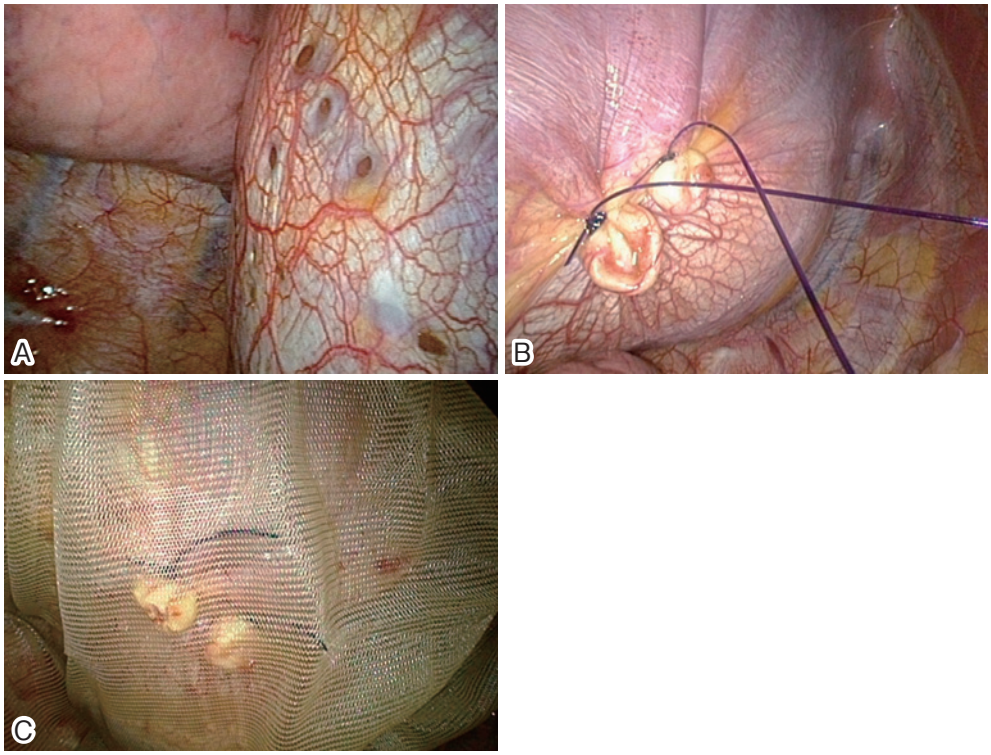

Fig. 1 Intraoperative findings in Case 1
The perforated holes were recognized on the diaphragm (A). Perforated sites were ligated and the diaphragm was reefed using an endoscopic loop (B). After reefing the tendon of the diaphragm by ligation, the diaphragm was covered with a PGA sheet (C).

operative findings (recurrent pneumothorax in synchrony with menses) and intraoperative findings ("blueberry spots," ie, endometrial gland or perforated holes on the diaphragm).

Preoperative and postoperative clinical findings (including age, laterality, recurrence) and surgical procedure, with or without chemical pleurodesis by adhesive agent(s) were evaluated in these five patients.

The patients provided prior written informed consent for inclusion in this report.

Treatment

With the patient under general anesthesia, VATS was performed with double-lumen endobronchial intubation, using one port site for the camera and a 5-cm mini-thoracotomy. Our surgical basic procedure (BP) was as follows.

Perforated sites or blueberry spots detected on the diaphragm (**Fig. 1A**) were ligated with an ENDOLOOP® endoscopic loop (Ethicon, Inc., Somerville, NJ, USA) (**Fig. 1 B**). After reefing the tendon of the diaphragm by ligation, the diaphragm was covered with a Vicryl Mesh® (Ethicon, Inc.) or a NEOVEIL Sheet® (Gunze Medical Ltd., Osaka, Japan) PGA sheet (**Fig. 1C**).

Three methods were used for the five patients: (A) BP only (without any adhesive chemical agent) in one case, (B) BP with tetracycline plus OK432 (an anti-cancer agent) in two cases, (C) BP with tetracycline only in two cases (because, on the recommendation of the Ministry of Health, Labor and Welfare, the use of OK432 has not been allowed by our Institutional Review Borad rules since 2019 for patients with benign disease).

We evaluated preoperative and postoperative clinical findings (including age, laterality, recurrence) and surgical procedures with or without chemical pleurodesis by adhesive agent(s) for these five patients.

Results

Case 1 (Procedure A)

A 33-year-old woman taking low-dose pills for dysmenorrhea was referred to our hospital for two episodes of right-sided SP. Both times, SP coincided with menstruation, and CP was strongly suspected. A preoperative chest CT scan revealed tiny bullae at the apex and S9 of the right lung. She opted for surgery. Intraoperative examination revealed bullae at the apex of the lung and perforation holes of the diaphragm. In addition to resection of the bullae, BP (reefing of the diaphragm by ligation and covering with a PGA sheet) was performed (3

mL of fibrin glue was sprayed over a PGA sheet). The duration of surgery was 95 minutes. The thoracic drain was removed on POD 2 and the patient was discharged. After discharge, low-dose pill therapy was resumed, and no recurrence has been observed for approximately 7 years after surgery.

Case 2 (Procedure B)

A 45-year-old woman was referred to our department for three episodes of right pneumothorax, all of which occurred during her menstrual period. CP was strongly suspected and she decided to undergo surgery. Preoperative chest CT revealed tiny bullae at the apex of the right lung. Intraoperatively, blueberry spots were found on the diaphragm. In addition to resection of the pleural apex, BP was performed and 200 mg of tetracycline and 2KE of OK432 were dissolved in 10 mL of saline and sprayed on a PGA sheet. The duration of surgery was 70 minutes. The thoracic drain was removed on POD 3 and the patient was discharged. No recurrence has been observed for 6 years postoperatively.

Case 3 (Procedure B)

A 34-year-old woman underwent surgery in the Department of Gynecology at our hospital for uterine myoma. She was referred to our department for postoperative development of right pneumothorax. She previously had chest pain and mild dyspnea during her menstrual periods, so CP was suspected and she decided to undergo surgery. Preoperative chest CT revealed pleural irregularities of the right pulmonary apex. Intraoperatively, we observed pleural irregularities of the apex and blueberry spots on the diaphragm. In addition to resection of the pleural apex, BP was performed. Then, adhesive therapy was performed with tetracycline and OK 432, as in Case 2. The duration of surgery was 110 minutes. The thoracic drain was removed on POD 3, and the patient was discharged. Histopathology revealed no emphysematous change in the resected lung. She conceived through artificial insemination after surgery and experienced a recurrence at 3 years after parturition (5 years after surgery).

Case 4 (Procedure C)

A 39-year-old woman previously experienced self-limiting chest pain and mild dyspnea during menstruation. On the second day of menstruation, she suddenly developed severe chest pain and dyspnea and was referred to our department with a diagnosis of right pneumothorax. She was admitted to our hospital, and a chest CT scan revealed bullae at the apex of the right lung. After thoracic drainage, she was discharged. However, 3 months later, she developed a similar clinical manifestation during menstruation, and was re-hospitalized with a diagnosis of recurrent right pneumothorax. CP was suspected and surgery was performed at her request.

Intraoperatively, blueberry spots were found on the diaphragm. In addition to resection of the pleural apex, BP was performed. Then, 200 mg of tetracycline was dissolved in 10 mL of saline and sprayed on a PGA sheet. The duration of surgery was 68 minutes. The thoracic drain was removed on POD 2 and the patient was discharged. No recurrence has been observed for 3 years postoperatively.

Case 5 (Procedure C)

A 42-year-old woman, developed dysmenorrhea and was twice referred to our department for right pneumothorax. Both episodes occurred during menstruation, and CP was suspected. She developed sudden-onset right chest pain and respiratory distress and was transported to our emergency center. Because the right lung had completely collapsed, a thoracic drain was inserted, and she was transferred to our department the next day. A chest CT scan revealed bullae at the apex and S6 of the right lung. Because air leakage persisted for longer than a week, surgery was performed for suspected CP.

Intraoperative examination revealed perforated holes on the diaphragm and perforated bulla on S6 of the lung. In addition to resection of the bullae (at the apex and S6), BP was performed after which adhesive therapy was performed with tetracycline as in Case4. The duration of surgery was 83 minutes. The thoracic drain was removed on POD 3 and the patient was discharged.

One month after surgery, at her next menstruation, she visited our outpatient clinic for follow-up, and a chest X-ray revealed mild collapse of the right lung, indicating postoperative recurrence. She was referred to the gynecology unit, where she was placed on hormonal therapy (dienogest) to prevent recurrence, including treatment of dysmenorrhea. No recurrence has been observed during the past 9 months.

Summary of Results

The ages of the patients ranged from 33 to 45 years (mean, 38.6±5.1 years). CP occurred on the right side in all cases. Partial lung resection/bullectomy was performed in all cases. The mean duration of surgery was 85.2±17.6 minutes (range, 68-110 minutes). In four of five cases, histopathology revealed emphysematous change in the resected lung. Recurrence developed in two of the five cases (one each for procedures B and C) (**Table 1**).

Table 1 Patients' characteristics and outcome

	Case 1	Case 2	Case 3	Case 4	Case 5	Mean
Age (y/o)	33	45	34	39	42	38.6
Laterality	Right	Right	Right	Right	Right	
Bullae/blebs on preoperative CT	Apex+S9	Apex	None	Apex	S6	
Diaphragm findings	Perforated holes	Blueberry spots	Blueberry spots	Blueberry spots	Perforated holes	
Lung resection	Apex+S9	Apex	Apex	Apex	Apex+S6	
Adhesive agents TC/OK432	A: none (fibrin glue)	B: 200 mg/ 2KE	B: 200 mg/ 2KE	C: 200 mg/-	C: 200 mg/-	
Duration of Surgery (min.)	95	70	110	68	83	85.2
Drain discharged on	POD2	POD3	POD3	POD2	POD3	2.6
Recurrence	-	-	5 years after	-	1 month after	
Hormonal therapy after surgery	Low-dose pill	None	None, but pregnancy after surgery	None	Dienogest	
Pathology of resected lung	Emphysematous bulla	Emphysematous bulla	No emphysema	Emphysematous bulla	Emphysematous bulla	
Others (preoperative)	Low-dose pill for dysmenorrhea	None	Surgery for uterine myoma	None	Dysmenorrhea	

Discussion

CP is the most common manifestation of TES, as endometrial glands are present in the thoracic cavity, in the lung parenchyma, and on the diaphragm. CP is defined as SP recurrence within 72 hours of menstruation onset[1]. Previously, SP due to CP was believed to be rare. Nakamura et al.[2] reported that in 1984 CP was diagnosed in less than 1% of women with SP in Japan. However, some recent studies reported that SP was caused in 24.6-29.0% of women with SP[3–5]. The increased rate of diagnosis of SP in women may be attributable to increased understanding of CP among physicians.

CP can be treated by hormonal therapy. Low-dose pills or dienogest is used for dysmenorrhea. Although both inhibit endometrial proliferation, it is difficult to control recurrence with hormonal therapy alone. Surgical treatment for CP includes laser ablation and partial pulmonary resection (if endometrial glands are in the lungs) by VATS. When endometrial glands are present on the diaphragm, as in our patients, the conventional surgical procedure is resection of the diaphragm, including the endometrial glands, and reconstruction with a synthetic mesh by VATS and/or video-laparoscopic surgery. In addition, intraoperative adhesive therapy with talc or tetracycline decreases the rate of postoperative recurrence, as compared with surgery alone. However, in a review of 10 studies, postoperative recurrence occurred in 29.0% (139/478) of cases. These results were not stratified by surgical procedure or postoperative hormonal treatment[6]. A review article reported that postoperative hormonal treatment reduced CP recurrence rates[7].

Previously, we described the use of diaphragmatic reefing with ENDOLOOP® ligation, PGA sheet covering, and intraoperative adhesion therapy for four patients with hydrothorax due to pleuroperitoneal communication (PPC), which can occur in patients undergoing continuous ambulatory peritoneal dialysis. There were no cases of recurrence[8]. In the present study, a similar technique was performed for CP. The conventional surgical procedure for a diaphragmatic lesion (ie, diaphragmatic resection and reconstruction) is associated with a higher risk of complications, such as postoperative pleural effusion, diaphragmatic hernia due to suture failure, and infection caused by foreign objects. As compared with the conventional procedure, our technique seems to be simpler and safer. CP recurred in two of the five present patients; however, the recurrence rate will likely be lower in patients receiving postoperative hormonal treatment.

Conclusions

We treated five cases with CP (10% of all SP among women) during a recent 10-years periods. Although the small number of patients is a limitation of our study, our experience suggests that surgery is the best treatment for CP, along with adhesion therapy and postoperative hormonal therapy to prevent CP recurrence. With respect to

surgical technique, our procedure (diaphragmatic reefing by ligation and covering with a PGA sheet) seems to be simpler and more effective than the conventional procedure (resection and reconstruction with a synthetic mesh of the diaphragm).

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