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Surgical Treatment of Catamenial Pneumothorax

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OBJECTIVE: To discuss the aetiology and determine the optimal surgical treatment of catamenial pneumothorax.
METHODS: Between January 1980 and December 2007, 17 patients with catamenial pneumothorax were treated at our institution. Regarding the surgical approach, thoracotomy was performed until 1991, and thoracoscopic surgery was performed from 1992 onward.
RESULTS: Pneumothorax was on the right side in all but two patients. Surgery was performed on 15 of the 17 patients. The surgical procedure was a diaphragm resection plus a partial bleb resection in eight patients, a diaphragm resection in two patients, a diaphragm resection plus an absorbable polyglycolic acid sheet in four patients, and a partial bleb resection in one patient. Five patients demonstrated a postoperative recurrence (33.0%). However, no recurrence has been observed thus far in the four patients with an absorbable polyglycolic sheet placed on the diaphragmatic surface.
CONCLUSION: Although the postoperative recurrence rate of patients undergoing surgical procedures remains high, there were some patients with no postoperative recurrence. Placement of an absorbable sheet on the diaphragmatic surface may therefore prevent recurrence of catamenial pneumothorax. [*Asian J Surg* 2010;33(4):199–202]

Key Words: absorbable polyglycolic acid sheet, catamenial pneumothorax, thoracoscopic surgery

Introduction

Catamenial pneumothorax (CP) is currently considered to be a very unusual clinical condition. This entity exhibits a temporal relationship between the onset of pneumothorax symptoms and menstruation. The pneumothorax tends to occur on the right side in many patients, and is said to be accompanied by perforations and/or nodules in the tendinous part of the diaphragm. However, no definitive treatment for this condition has been established. Therefore, in this study, we retrospectively investigated the aetiology and attempted to determine the optimal surgical treatment for CP.

Patients and methods

Of the 215 female patients hospitalized and treated at Itabashi Hospital of Nihon University (Tokyo) between January 1980 and December 2007, 17 (7.9%) were classified as having CP. The median age of the patients at the time of operation was 41 years (range, 31–50 years). CP was defined as recurrent pneumothorax (at least two

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episodes) occurring between the day before and within 72 hours after the onset of menstruation. In some cases, the cancer antigen 125 (CA-125) blood level was measured at the time of hospitalization. For the surgical approach, thoracotomy was performed until 1991, and thoracoscopic surgery was performed from 1992 onward.

In patients with a diaphragmatic abnormality, a partial resection of the diaphragm and a histopathological examination were performed. The surgical procedure was a diaphragm resection plus a partial bleb resection in eight patients, a diaphragm resection in two patients, and a partial bleb resection in one patient. After diaphragmatic resection in the thoracotomy procedures, we performed the additional process of placing continuous sutures in the absorbable suture material. On the other hand, after diaphragmatic resection by an endoscopic surgical stapler in thoracoscopic surgery, a stapler and stapler for cross-site were reinforced by simple interrupted sutures. Furthermore, the last four patients had an absorbable polyglycolic acid sheet (Neoveil, Gunze Ltd., Kyoto, Japan) placed on the diaphragmatic surface after diaphragm resection. The polyglycolic acid sheet was held in place by stitches at its periphery or biological glue. All surgical patients were interviewed by phone to obtain data on long-term outcomes before preparing this paper. The observation periods of the surgical patients ranged from 1 year (shortest) to 13 years (longest) (mean, 60.8 months). Informed consent was obtained from all patients participating in the study, and the research procedures were in accordance with the recommendations of the Helsinki Declaration of 1975 and its successive modifications.

Results

Pneumothorax was observed on the right side in all but two patients. Of the 17 patients, 2 did not wish to undergo surgery and were therefore recommended to undergo hormone therapy; the remaining 15 patients underwent surgery. The previous medical histories of the cases included one patient who had undergone a hysterectomy and unilateral ovariectomy, and one patient that was undergoing treatment for endometriosis of the uterus. For the surgical approach, thoracotomy was performed in 5 patients, and thoracoscopic surgery was performed in 10 patients. Diaphragmatic abnormalities (Figure 1) included perforations and/or nodules in 14 of the 15 patients (93.3%) that underwent surgery (Table 1). Endometrial tissue (Figure 2) was found in the resected diaphragm in four patients (26.7%), and evidence of oestrogen receptor (+) and progesterone receptor (+) tissue was observed in another patient (6.7%) via immunostaining. The CA-125 level was measured 12 times each in 10 cases and ranged from 20.7 to 941.8 IU/mL (mean, 124.2 IU/mL); the level was within the normal range (35 IU/mL or less) in five patients. Five patients demonstrated postoperative recurrences (33.0%) out of all surgical patients. However, while the observation period was

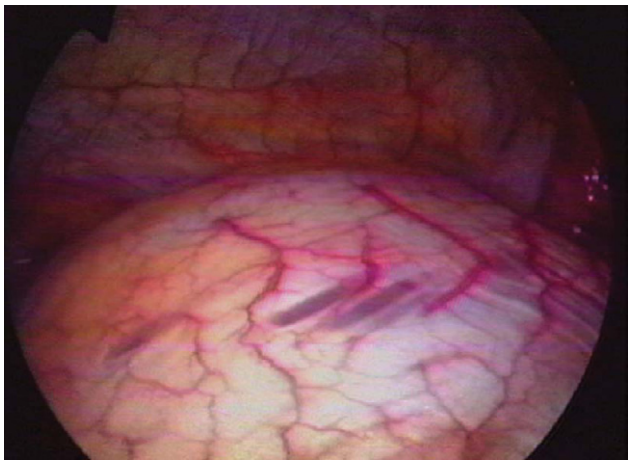

Figure 1. Intraoperative view of the tendinous part of the right diaphragm. Multiple perforations were found, and the convex apical surface of the liver can be observed.

Table 1. Intraoperative findings, surgical procedures and recurrence rate ($n = 15$)

Diaphragmatic abnormality*	Patients (n)	Procedures	Recurrence (%)
Diaphragmatic abnormality (+) Blebs/bullae (+)	10	DR or DR + bullectomy	4 (40%)
Diaphragmatic abnormality (+) Blebs/bullae (−)	4	DR + PGA	0 (0%)
Diaphragmatic abnormality (−) Blebs/bullae (+)	1	Bullectomy	1 (100%)

*Diaphragmatic abnormality refers to perforations and/or nodules. DR = diaphragm resecion; PGA = polyglycolic acid sheet (Neoveil).

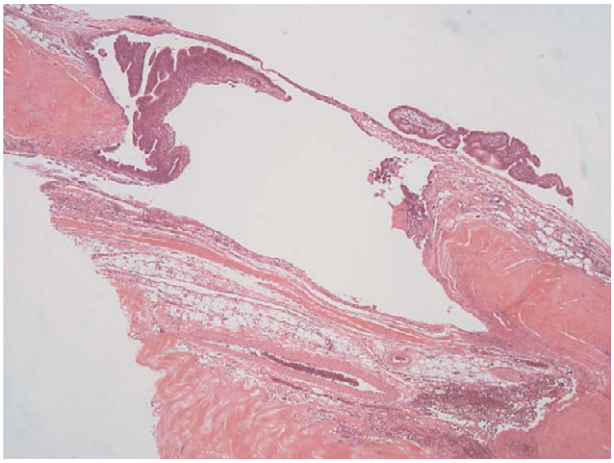

Figure 2. Section of the diaphragm shows an endometriotic focus composed of both glands and stroma within the diaphragm (haematoxylin and eosin, 40×).

short, no recurrence has so far been observed in the four patients with an absorbable polyglycolic sheet on the diaphragmatic surface (Table 1). For the five patients with postoperative recurrence, hormone therapy was provided for a minimum of 6 months.

Discussion

CP was first reported by Maurer et al,[1] and is a form of ectopic endometriosis that was named by Lillington.[2] Specifically, CP is a recurrent pneumothorax that occurs in concurrence with the menstrual cycle, often on the right side, and is said to involve diaphragmatic abnormalities but no obvious subpleural blebs, such as those observed in typical cases of spontaneous pneumothorax. However, it remains difficult to prove the presence of endometrial tissue in the pleural space, and, even when only a diaphragmatic abnormality is observed, all cases of recurrent pneumothorax that occur in concurrence with the menstrual cycle are considered cases of CP. In recent years, there have been increasing numbers of reports of CP. This increase may be due to the adoption of thoracoscopes that allow for examination of the diaphragmatic surface, which could not be observed in detail through a thoracotomic approach, or to the ease of performing thoracoscopy due to the use of minimally invasive procedures. Many of these cases occur on the right side, and perforations and/or nodules (blueberry spots) have reportedly been observed in the tendinous diaphragm. In fact, in slightly more than 90% of our surgical cases, we were able to confirm the presence of diaphragmatic abnormalities.

Regarding the mechanisms of CP development, several hypotheses have been suggested. The first hypothesis is related to diaphragmatic perforations caused by endometriosis and airflow from the genital tract into the chest.[1,3] The second hypothesis is related to endometrial implants in the visceral pleura.[4] The third hypothesis is related to the level of prostaglandin F2α in the blood.[5] In many cases, no abnormalities are observed in the visceral pleural, whereas perforations are observed in the diaphragm, and pneumothorax can be induced through pneumoperitoneum. Thus, the first hypothesis (the mechanism involving the transdiaphragmatic passage of air) is currently the most favoured; however, it cannot explain the pathogenesis of all the reported cases of CP because there are also reports in which pneumothorax could not be induced by pneumoperitoneum[1] and reports of CP occurring after a hysterectomy.[6]

Furthermore, hypotheses related to hematogenous[7] or lymphogenous[8,9] dissemination have been offered to explain the route of the ectopic endometrial tissue to the pleural space. Many reports discuss the intrathoracic dissemination of endometrial tissue interposed between perforations in the diaphragm, and pneumothorax predominates on the right side, thus leading us to believe that the hypotheses of transdiaphragmatic dissemination are the most compelling.

There are cases in which the CA-125 blood level is measured before surgery to provide an auxiliary diagnosis. However, CA-125 levels only reflect the amount of endometrial tissue, and it is currently believed that the usefulness of such measurements is low.

In investigating the best and most effective treatment for CP, we found that earlier reports recommended performing hormone therapy for approximately 6 months; if this proved to be ineffective, then surgical procedures, such as diaphragm resection and pleurodesis, were performed.[4,10,11] However, surgical procedures were recently performed for diagnosis and treatment and followed by approximately 6 months of hormone therapy in cases in which the presence of ectopic endometrium had been proven.[12] The recurrence rate after performing these surgical procedures alone is less than 30%.[13] In contrast, the recurrence rate after hormone therapy alone is more than 50%.[13] Additionally, the long-term continuation of this therapy is also very difficult due to the onset of various side effects.

Therefore, the results of hormone therapy alone are poor compared with the results of performing surgery alone to treat the diaphragmatic abnormalities. This suggests

that for effective treatment, it is important to surgically locate and treat the diaphragmatic abnormalities without fail. This surgical reduction of endometrial tissue with diaphragm resection and the interruption of the transperitoneal-transdiaphragmatic migration of endometrial tissue may thus provide good results. Furthermore, in our one patient, no diaphragmatic abnormalities were observed at the time of the operation, but some diaphragmatic abnormalities (nodules) were observed at the time of postoperative recurrence. The timing of the operation is, therefore, considered to be an important factor in identifying the presence of abnormalities in the thoracic cavity in patients presenting with CP.

In the future, after excluding typical cases of pneumothorax (rupture of subpleural blebs) from cases of CP, we intend to consider the surgical period and to perform either a partial resection while covering the surgical site with an absorbable polyglycolic acid sheet in cases involving an abnormality on the diaphragmatic surface, or cover the site with an absorbable polyglycolic acid sheet in cases in which no abnormalities are observed on the diaphragmatic surface.

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