Translated by E.V. Trushina

Research Article

https://doi.org/10.23934/2223-9022-2024-13-3-399-409

Treatment Strategy for Catamenial and Endometriosis-Related Recurrent Spontaneous Pneumothorax

O.V. Voskresensky[1, 2] ✉, E.A. Tarabrin[1, 3], M.M. Damirov[1], P.A. Yartsev[1], R.N. Chirkov[2]

Scientific Department of Emergency Surgery, Endoscopy and Intensive Care
[1] N.V. Sklifosovsky Research Institute for Emergency Medicine
Bolshaya Sukharevskaya Sq. 3, Moscow, Russian Federation 129090
[2] Tver State Medical University
Sovetskaya Str. 4, Tver, Russian Federation 170100
[3] I.M. Sechenov First Moscow State Medical University
Trubetskaya Str. 8, bldg. 2, Moscow, Russian Federation 119048

✉ **Contacts:** Oleg V. Voskresenskiy, Doctor of Medical Sciences, Surgeon, Senior Lecturer, Training Center, N.V. Sklifosovsky Research Institute for Emergency Medicine.
Email: oleg_vskr@mail.ru

RELEVANCE The problem of treating thoracic endometriosis complicated by recurrent spontaneous catamenial pneumothorax is associated with the widespread prevalence of endometriosis, the complexity of its treatment and unsatisfactory long-term outcomes. The diagnosis of catamenial pneumothorax can only be established with a carefully collected history and confirmation of its recurrent nature. Favorable results are achieved with complex therapy carried out by the thoracic surgeon and gynecologist, consisting of an adequate volume of surgical intervention and hormonal therapy with gonadotropin releasing hormone agonists.

AIM OF STUDY To improve the diagnosis of thoracic endometriosis and recurrent catamenial and endometriosis-related spontaneous pneumothorax. To optimize the currently available treatment tactics by reducing the incidence of early relapses of catamenial pneumothorax and creating favorable conditions for the formation of reliable pleurodesis.

MATERIAL AND METHODS We studied the immediate outcomes of complex treatment of 30 patients (14.7%) with right-sided recurrent catamenial (28) and endometriosis-related spontaneous pneumothorax (2), with a median age of 41 (37;44) years, for the period from 2011 to the first half of 2023. Surgical treatment via video-assisted thoracoscopic access was performed in 26 patients (86.7%). After surgery, all the patients were recommended a six-month course of hormonal therapy with gonadotropin releasing hormone agonists.

RESULTS Various types of mechanical pleurodesis were performed in 25 patients (96.1%), resection of the diaphragm — in 17 (65.4%), alloplastic reconstruction with a synthetic mesh implant — in 12 patients (46.1%), pulmonary resection — in 9 (34.6%). In 12 of 26 patients (46.1%) in the early postoperative period, 6 (3; 6.75) days after surgery, a recurrence of catamenial pneumothorax developed, which subsequently required repeated interventions. In 5 patients, the relapse clearly coincided with the menstrual cycle. Early recurrence of pneumothorax required repeated drainage of the pleural cavity in all the patients, chemical pleurodesis in 2, and prolonged drainage of the pleural cavity in 1.

CONCLUSION Surgeries involving resection and reconstruction of the diaphragm with a mesh implant, pleurectomy, lung resection do not ensure the removal of all endometrioid heterotopias of the diaphragm and lung, therefore, during the first mensis after surgery, in the early postoperative period, 46.1% of the patients had a recurrence of catamenial pneumothorax, with no formed pleurodesis, which contributes to the possibility of further recurrences of pneumothorax. Improving the immediate outcomes of complex treatment of catamenial pneumothorax is possible with the preventive administration of hormonal therapy 1.5–2 months before the planned surgical intervention, which requires a multidisciplinary approach of the thoracic surgeon and gynecologist.

Keywords: thoracic endometriosis, catamenial pneumothorax, surgical treatment, immediate outcomes

For citation Voskresenskiy OV, Tarabrin EA, Damirov MM, Yartsev PA, Chirkov RN. Treatment Strategy for Catamenial and Endometriosis-Related Recurrent Spontaneous Pneumothorax. *Russian Sklifosovsky Journal of Emergency Medical Care*. 2024;13(3):399–409. https://doi.org/10.23934/2223-9022-2024-13-3-399-409 (in Russ.)

Conflict of interest Authors declare lack of the conflicts of interests

Acknowledgments, sponsorship The study has no sponsorship

Affiliations

Oleg V. Voskresensky	Doctor of Medical Sciences, Surgeon, Senior Lecturer, Training Center, N.V. Sklifosovsky Research Institute for Emergency Medicine; Associate Professor, Department of Oncology, Surgery and Palliative Medicine, Tver State Medical University; https://orcid.org/0000-0003-3018-3517, oleg_vskr@mail.ru; 40%, surgery, collection and analysis of material, collection and analysis of literature data, text writing
Evgeniy A. Tarabrin	Doctor of Medical Sciences, Leading Researcher, Scientific Department of Emergency Surgery, Endoscopy and Intensive Care, N.V. Sklifosovsky Research Institute for Emergency Medicine; Head, Department of Hospital Surgery No. 2, N.V. Sklifosovsky Institute of Clinical Medicine, I.M. Sechenov First Moscow State Medical University; Director, Clinic of Hospital Surgery No. 2; https://orcid.org/0000-0002-9616-1161, t_evg_a@mail.ru; 30%, surgery, editing the article on thoracic surgery issues

Mikhail M. Damirov	Doctor of Medical Sciences, Full Professor, Head, Scientific Department of Emergency Gynecological Diseases, N.V. Sklifosovsky Research Institute for Emergency Medicine; https://orcid.org/0000-0001-6289-8141, damirov@inbox.ru; 15%, consulting patients in the study group, determining indications for prescribing hormonal therapy, editing the article on endometriosis treatment issues
Petr A. Yartsev	Doctor of Medical Sciences, Full Professor, Head, Scientific Department of Emergency Surgery, Endoscopy and Intensive Care, N.V. Sklifosovsky Research Institute for Emergency Medicine; https://orcid.org/0000-0003-1270-5414, 79262265893@yandex.ru; 10%, editing the text of the article
Roman N. Chirkov	Doctor of Medical Sciences, Full Professor, Head, Department of Oncology, Surgery and Palliative Medicine, Tver State Medical University; https://orcid.org/0000-0002-9606-7351, tak.tgmu@mail.ru; 5%, selection of literature, general editing

CP — catamenial pneumothorax
ERP — endometriosis-related pneumothorax
GnRH — gonadotropin-releasing hormone
IHC — immunohistochemistry
PSP — primary spontaneous pneumothorax
TE — thoracic endometriosis

INTRODUCTION

Endometriosis, as follows from the Clinical Guidelines approved by the Ministry of Health of the Russian Federation in 2020, is a pathological process in which tissue similar to the endometrium in its morphological and functional properties is detected outside the uterine cavity. This disease affects approximately 10% of women of reproductive age worldwide. The features of this benign disease are as follows: the ability to infiltrative growth, a tendency to recurrence, and invasive growth. There are two directions of endometriosis treatment — hormonal therapy and surgical interventions [1]. Damage to organs and structures of the pleural cavity — thoracic endometriosis (TE) — is not the most common localization of extragenital endometriosis. It is not listed in a separate category in ICD-10, but belongs to categories N80.8 — “Other endometriosis” or N80.9 — “Endometriosis, unspecified”. But in ICD-11, it is already coded as: “GA10.G - Endometriosis of the chest: XK7F - Superficial, XK16 - deep.”

Catamenial pneumothorax (CP) is a spontaneous pneumothorax that develops 24 hours before and within 72 hours after the onset of mensis. Endometriosis-related pneumothorax develops against the background of TE, but is not associated with the menstrual cycle [2]. CP is the most common manifestation of TE, reaching 73%. CP is a repeatedly recurring disease [3]. Exclusively surgical treatment of CP is not a reliable method. The recurrence rate of endometriosis-related pneumothorax after surgical treatment is 27.5–40% [4–6]. Recurrence of endometriosis-related pneumothorax (ERP) was 27%, whereas in case of non-catamenial and non-endometriosis-related pneumothorax the rate of postoperative recurrence was 5.3% over an observation period of 32.7 months [5]. Thus, CP and ERP are a complex and urgent problem that requires clarification of the optimal treatment tactics.

The aim of the study was to reduce the incidence of CP relapses after surgical treatment; to analyze the results of intraoperative diagnosis of TE and to evaluate the immediate outcomes of surgical treatment of TE complicated by CP; to develop treatment tactics aimed at improving the treatment outcomes of patients with CP.

MATERIAL AND METHODS

From 2011 to June 2023, 847 patients with the diagnosis of "spontaneous pneumothorax" were treated at the N.V. Sklifosovsky Research Institute for Emergency Medicine. There were 204 women (24.1%) with a mean age of 38.9±9.5 years. According to the anamnesis, 28 patients had CP, and 2 patients had ERP. Thus, pneumothorax that developed against the background of TE was detected in 30 of 204 patients, which amounted to 14.7%. The median age of patients with CP and ERP was 41 (37; 44) years, ranging from 25 to 51 years. All the patients had right-sided pneumothorax, which is fully consistent with the literature data [7; 8]. Eight patients were hospitalized with the first episode of pneumothorax; recurrent pneumothorax was detected in 22 of 30 patients (73.3%). The median incidence of radiographically recorded recurrent pneumothorax was 2 (1; 2), and the maximum number of episodes was 8. A total of 26 patients were admitted for

emergency indications, and 4 patients were admitted on a planned basis. Pleural drainage on admission was performed in 17 patients (65.4%). In nine patients (30%), drainage was not performed due to the small volume of pneumothorax and the absence of respiratory failure. Twenty-six of 30 patients (86.6%) underwent thoracoscopic surgery. In our publications of 2014–2015, we formulated the following tactics: in case of recurrent CP confirmed by anamnesis and radiological examinations, indications for surgical treatment were established, the scope of which depended on intraoperative findings. Video-assisted thoracoscopic access was performed. The diagnostic criterion for TE was the presence of fenestrations of the central tendon of the diaphragm, as well as endometrioid heterotopias in the form of brown spots or blue or violet formations, cyst-like red formations, brown deposits on the diaphragm or parietal pleura, diaphragm porosity zones (Fig. 1). Resection of the pathologically altered central tendon of the diaphragm with removal of visible pathological formations using an endoscopic suturing device was performed in 16 patients, and "manually" in 1 patient. In the absence of diaphragmatic perforations, the diaphragm was not resected but abraded to create pleurodesis between the basal segments of the lung and the diaphragm (Fig. 2). When focal or emphysema-like changes in lung tissue were detected, a sparing marginal resection of the altered area of the lung was performed. Parietal pleurectomy or pleural abrasion to "blood dew" was performed (Fig. 3) [8; 9]. Later, the diaphragm abrasion was replaced by reconstructive surgery with polypropylene mesh (measuring 15×15 cm) covering most of the diaphragm surface. The mesh was fixed with interrupted sutures, both at the edges and in the central section of the diaphragm, with interrupted prolene sutures through the stapled suture of the diaphragm.

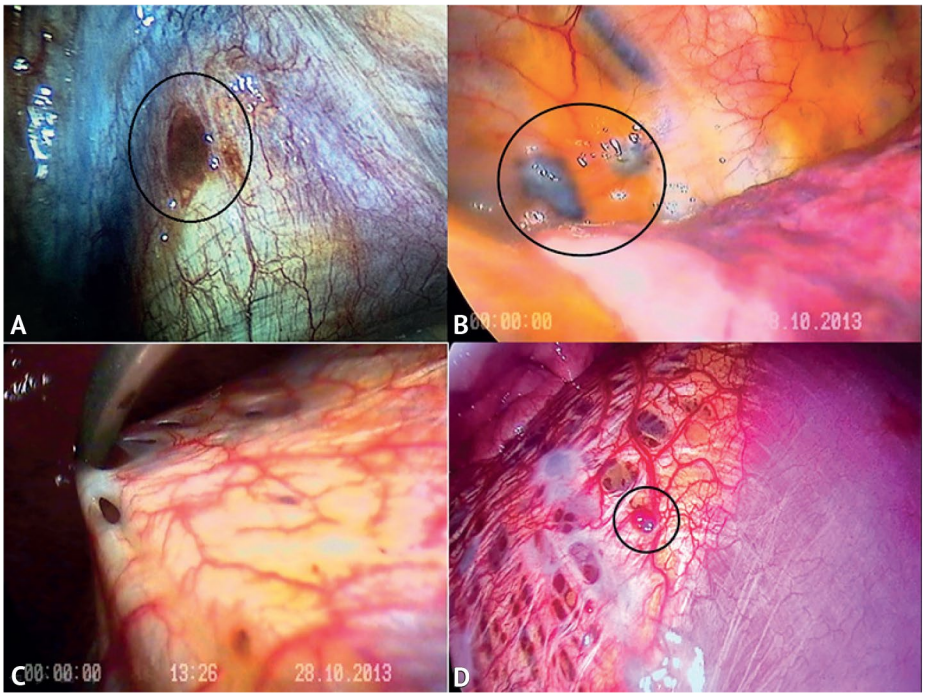

Fig. 1. Endometrioid heterotopias and fenestrations of the diaphragm and parietal pleura. A — brown spot on the diaphragmatic peritoneum (circled) revealed during transdiaphragmatic laparoscopy; B — endometrioid heterotopia in the form of a purple nodule on the parietal pleura (circled); C — pass-through endometrioid fenestrations of the tendinous center of the diaphragm; D — zone of widespread porosity of the diaphragm (marked with arrows) and endometrioid heterotopia of the diaphragm in the form of a red cystic formation (circled), brown plaque on the diaphragm

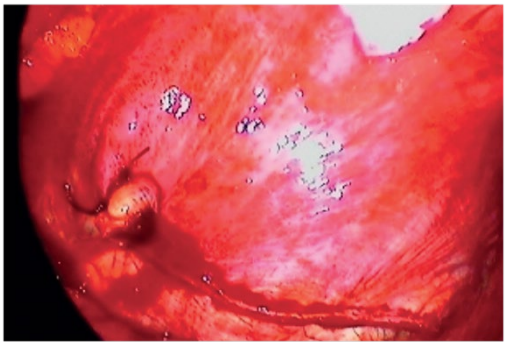

Fig. 2. Endophoto. The result of pleural abrasion of the diaphragm to the "blood dew"

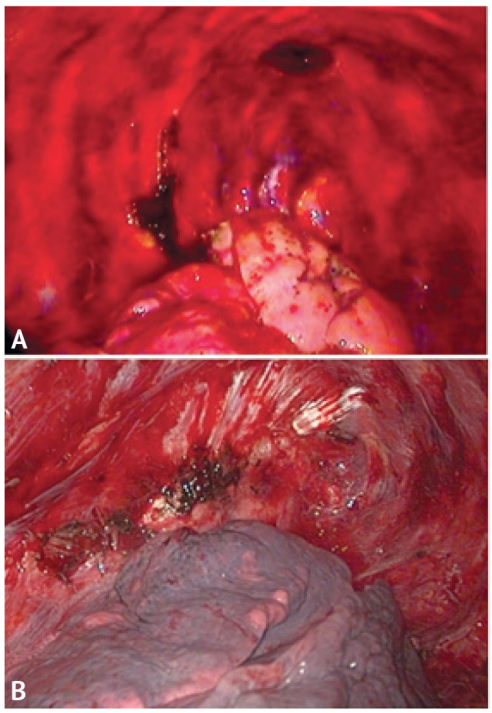

Fig. 3. Pleurectomy and pleural abrasion. A — Subtotal pleurectomy; B — pleural abrasion (polypropylene mesh abrasion)

Patients with a history of adenomyosis or endometriosis were examined by the gynecologist before surgery to determine the localization and stage of the process. After surgery, all the patients with detected signs of TE were re-consulted by the gynecologist to prescribe hormonal therapy. A 6-month course of treatment with gonadotropin-releasing hormone (GnRH) agonists was recommended as the primary therapy for endometriosis. The drug was administered on the 2nd day of the first menstruation after surgery.

In case of early recurrence of CP (in the period from the moment of surgery until discharge), repeated drainage of the pneumothorax zone was performed and, according to indications, chemical pleurodesis with povidone-iodine or doxycycline was performed [10; 11].

RESULTS AND DISCUSSION

The clinical picture of TE and the pneumothorax that developed against its background was manifested by pain in the right hemithorax, which arose the day before or on the 1st–2nd day of mensis in 24 of our patients (76.6%). The frequency of pain forced 5 women (19.2%) to consult a neurologist and receive treatment for dorsopathy and intercostal neuralgia. According to the literature, pain in the scapula or chest during mensis is highly specific for TE [2]. A complete gynecological history and complaints characteristic of TE provide an opportunity to correctly navigate the specifics of the disease. This allowed a group of Japanese authors to develop an assessment scale based on the most obvious clinical manifestations of TE, which can be used to speak about the presence of CP with a high probability of prognosis [12; 13]. Other symptoms – dyspnea in 16 patients (61.5%), discomfort, shortness of breath when inhaling, heaviness in the chest – were much less common and not specific for CP. In the absence of clinical manifestations, CP was diagnosed in 2 patients during a medical examination. According to the literature, chest pain at the beginning of mensis is typical for 90% of patients with CP, while respiratory distress occurred in 31% of patients. In 90% of women, symptoms of right-sided CP occurred on the 2nd day of menstruation [14]. Comparison of the obtained results with literary data showed a high degree of correspondence between the clinical manifestations in our group of patients and literary data.

A history of adenomyosis and/or external genital endometriosis was present in 14 patients (53.8%). Five of them had undergone various surgical interventions associated with this disease. In 2 patients, the only manifestation of pelvic endometriosis was regular lower abdominal pain on the first day of menstruation. In total, pelvic pain accompanying mensis was observed in 5 patients (19.2%). Secondary infertility was observed in 8 women (30.8%). Three of them participated in the in vitro fertilization program, but without a positive result. Two women became pregnant early in the

treatment of endometriosis with GnRH agonists, but endometriosis subsequently recurred, and repeated attempts to become pregnant were unsuccessful. There were no anamnesis data on gynecological diseases in 12 patients (46.1%). According to literary sources, external genital endometriosis was observed in 50–85% of patients with TE [7], which does not differ from the data of the present study. It was noted that in most patients, clinical manifestations of TE developed 5–7 years after the onset of symptoms of pelvic endometriosis [3]. Infertility and a history of uterine surgery are significantly associated with the development of CP or ERP [2]. From the above it follows that in patients of fertile age with pneumothorax, a thorough collection of anamnesis, including gynecological, is decisive in making the correct diagnosis.

The recurrent nature of CP is established if spontaneous pneumothorax occurs during mensis and is recorded by diagnostic radiology methods at least 2 times. Then it can be regarded as CP, and not as a random coincidence [5]. We have observations where in 2 women, who were subsequently diagnosed with fenestrations of the diaphragm, clinical manifestations of ERP occurred not during mensis, but during ovulation. The incidence of ERP occurring in the intermenstrual period, according to the literature, ranges from 37.9 to 63% [5, 15, 16]. Therefore, we differentiate the concepts of CP and ERP, in contrast to the position of our colleagues [17]. Non-catamenial ERP is much more difficult to detect when collecting anamnesis. In this regard, it is necessary to carefully study all observations of right-sided pneumothorax in the female population.

Videothoracoscopy is the only method of objective diagnostics of TE, allowing not only visualization, but also obtaining material for histological and immunohistochemical (IHC) analysis [18]. As can be seen from the table, in the studied group of patients, diaphragmatic fenestrations were present in 22 patients (84.6%). According to P. Bagan (2003), diaphragmatic defects were detected in 60% of patients with CP [19]. In addition to clearly defined fenestrations with a diameter of 5 mm or more, some authors pointed out small defects with sizes from 1 to 3 mm [20]. Purple spots or nodular formations were detected in 8 patients (30.8%). Formations resembling purple berries and purple spots, changes in the form of red and brown nodules characteristic of endometriosis were described in foreign literature [16]. In two of our observations, there was local fibrosis of the visceral pleura in 1 patient and of the parietal pleura in 2 patients. White nodular formations up to 5 mm in size in the in the S1 segment, fibrin deposits on the diaphragm and on the visceral pleura in the area of the apical segment - also in 1 patient. These changes were not previously associated with endometriosis in any way. However, according to foreign literature, white nodular formations up to 2 mm in size, localized on the visceral pleura of the apical segment, as well as small yellow nodules on the diaphragm, also had an endometrioid nature confirmed by the histological method [21]. Other authors have shown obvious signs of endometriosis, such as scars of the visceral and parietal pleura, funnel-shaped depressions on the parietal pleura, reaching the intercostal muscles in depth [16]. Changes on the surface of the diaphragm and visceral pleura were detected in 30–40% of patients with CP [3, 7]. At the same time, the absence of any findings was observed in 8.5% of patients [7].

In 4 patients of our group, small bullae up to 2 cm were detected on CT. In 6 operated patients (23.1%), blebs up to 1 cm in diameter were detected during revision. And only one patient was diagnosed with a 1.7 cm bulla. Blebs were localized in the 4th segment in 3 patients, in the 1st segment in 2 patients, and in one observation in the 3rd, 8th, 9th, and 6th segments, both along the edge of the segments and along the interlobar and costal surfaces. Marginal lung resection for biopsy was performed in 9 patients (34.6%). According to foreign literature, air cysts were more common in visceral endometriosis than other formations. The most frequent locations of endometriosis on the visceral pleura were the 4th, 6th, and 2nd segments of the lung. In this case, endometrioid foci were more often found in the zone of "contact" of the lobes — along the interlobular surfaces of the segments, as well as along the edge of the segments. Based on histological studies, a pathogenesis of visceral pleural endometriosis was proposed [16, 22, 23]. At the histological level, it was shown that endometrioid stroma was present in the bleb wall [22]. This study emphasizes the need for lung resection with blebs, both in terms of removing potential foci of endometriosis and histological verification of the disease. According to literary data, one of the main tasks during surgical intervention was considered to be the removal of all visible foci of endometriosis [24].

Table

Volume of surgical intervention in patients with endometriosis-related spontaneous pneumothorax, immediate and remote outcomes

Patients No.	Pleurodesis	Diaphragm fenestrations	Suturing of fenestrations	Diaphragm abrasion	Resection of the diaphragm	Diaphragmatic mesh repair	Lung resection	Relapse during hospitalization
1	CPE	Yes	–	–	–	–	Yes	Yes
2	A	Yes	–	Yes	Yes	–	–	–
3	No	Yes	Yes	–	–	–	–	–
4	A	Yes	Yes	Yes	–	–	Yes	–
5	CPE +A	Yes		Yes	Yes	–	Yes	–
6	SPE	Yes		Yes	Yes	–	–	–
7	CPE	Yes	–	–	–	–	–	–
8	CPE	Yes	–	–	–	–	–	–
9	CPE	Yes	–	–	Yes	Yes	–	Yes
10	CPE	Yes	–	–	Yes	Yes	Yes	–
11	CPE	Yes	–	–	Yes	Yes	–	–
12	CPE	Yes	–	–	Yes	Yes	–	–
13	CPE	Yes	–	–	Yes	Yes	Yes	Yes
14	CPE	Yes	–	–	Yes	Yes	–	Yes
15	CPE	Yes	–	–	Yes	–	–	–
16	CPE	Yes	Yes	–	Yes	Yes	–	Yes
17	SPE	Yes	–	–	Yes	Yes	Yes	–
18	SPE	Yes	–	–	Yes	Yes	–	Yes
19	CPE	Yes	–	–	–	–	Yes	Yes
20	CPE	Yes	–	–	Yes	Yes	–	Yes
21	CPE	Yes	–	–	Yes	Yes	–	–
22	CPE	Yes	–	–	Yes	Yes	–	–
23	SPE	No	–	Yes	–	–	Yes	Yes
24	CPE	No	–	–	–	–	Yes	Yes
25	CPE	No	–	–	–	–	Yes	Yes
26	RCPE +A	No	–	–	Yes	Yes	–	Yes
Total	25 (96.1%)	22 (84.6%)	3 (11.5%)	5 (19.2%)	17 (63.4%)	12 (46.1%)	10 (38.5%)	12 (46.1%)

Notes: CPE – costal pleurectomy (from 2 to 8 ribs); SPE – subtotal pleurectomy; RCPE – repeat costal pleurectomy

Resection of the diaphragm is a surgical stage aimed at stopping the abdominopleural air flow through fenestrations and removing endometrioid heterotopia. As follows from Table 1, diaphragm resection was performed in 17 patients (65.4%). Corrective intervention on the diaphragm was not performed in 7 patients (26.9%). In the first observation, the patient was found to have diaphragmatic fenestrations during revision, the nature of which was unclear to the surgeon at that time. The patient subsequently refused repeated intervention. In two patients with multiple fenestrations distributed throughout the central tendon of the diaphragm, resection of the diaphragm and removal of all visible openings was impossible due to extensive damage to the central tendon of the diaphragm. We present the opinion of the authors, who are absolute experts and have the largest number of publications and references to publications on this topic: "...we believe that the use of endoscopic staplers for the diaphragm can be considered safe (from the point of view of the possibility of developing late hernias) only if they are used for small resections and (or) for closing small defects, while in other cases its use is not recommended" [24]. In one case, it was decided not to resect the diaphragm because the patient was postmenopausal. In three patients, no macroscopic fenestrations of the diaphragm were detected. Resection of the diaphragm is justified in the presence of endometrioid heterotopia or fenestrations. The lesions should be removed and the defects should be corrected [25]. However, there are publications that describe the use of synthetic mesh for fenestrations without resection of the diaphragm [26].

Fenestration suturing was performed in 3 patients (11.5%). According to the literature, this method of eliminating diaphragmatic defects was used in combination with diaphragm abrasion and complete pleurectomy [25]. Diaphragm abrasion was performed in 5 patients (3.8%). In 6 patients (4 with diaphragm fenestrations and 2 without fenestrations), there were no interventions on the diaphragm. It should be emphasized that careful removal of all endometriosis foci contributes to a longer relapse-free course of the disease in the future [16]. Obviously, it is impossible to reliably remove all endometrioid heterotopias during resection of the altered diaphragm. Therefore, instead of diaphragm abrasion, we began to perform diaphragm reconstruction with a lightweight polypropylene mesh measuring 15×15 cm, covering most of its surface. Polypropylene mesh diaphragm repair in combination with diaphragm resection was performed in 12 patients (46.1%). The mesh suturing serves not so much to strengthen the stapled suture after resection, but rather to create a framework for obtaining a dense fibrous layer due to mesh encapsulation, which was proven by the example of remote outcomes of inguinal hernia surgery [27]. According to the literature, a mesh in addition to suturing diaphragmatic defects was used in cases of microporosity of the diaphragm [19]. This is consistent with our observations, in which out of 4 patients (15.4%) with CP, in the absence of diaphragmatic fenestrations, in 2 observations a punctate rash was noted on the visceral surface of the diaphragm: in one patient - on a limited area of a small surface (Fig. 3A), in the other, fine-point changes were detected almost over the entire surface of the diaphragm (Fig. 3Б). The first patient (Fig. 3A) underwent diaphragm abrasion, the second (Fig. 3Б) — resection of the tendon center with meshplasty. The diagnosis of TE was confirmed in both patients. In the first observation, endometrioid heterotopias were detected during repeated histological analysis and IHC examination of the removed parietal pleura. In the second observation - in the preparation of the resected diaphragm using the optical microscopy method.

In our observations, the diagnosis of TE based on intraoperative biopsy materials of the diaphragm and parietal pleura was established in 9 out of 26 patients (34.6%), which corresponds to international data (22–37%) [20].

Mechanical pleurodesis was performed in 25 patients (96.1%). Parietal pleural abrasion using a polypropylene mesh (mash pleural abrasion) was performed in 2 patients. Intensive impact with a polypropylene mesh ball in the final result is almost no different from pleurectomy. Pleurectomy of varying extent was performed in 23 patients (88.5%) (Fig. 4). Mechanical pleurodesis was not performed in one patient. As follows from Table 1, costal pleurectomy (pleural resection from the II to the VIII rib level) was performed in 17 patients (65.4%); subtotal pleurectomy (removal of a larger volume of pleura, starting from the dome of the pleural cavity and up to the costophrenic sinus, and from the level of the sternum and up to the level of the thoracic sympathetic trunk) — in 4 cases (15.4%). According

to the literature, in CP for the purpose of pleurodesis, both abrasion of the parietal pleura, pleurectomy, and talc pleurodesis are used [5, 16, 19]. Subtotal pleurectomy was described as a method used for secondary pneumothorax both in the context of chronic obliterative pulmonary disease and in primary spontaneous pneumothorax in the absence of a substrate for pneumothorax in the lung or in the presence of persistent air flow through drainage tubes [28]. Being repeatedly recurring, CP, according to our data and the opinion of foreign authors, is long-term persistent [21]. Therefore, subtotal pleurectomy is the optimal option for mechanical pleurodesis, as evidenced by foreign authors too [26].

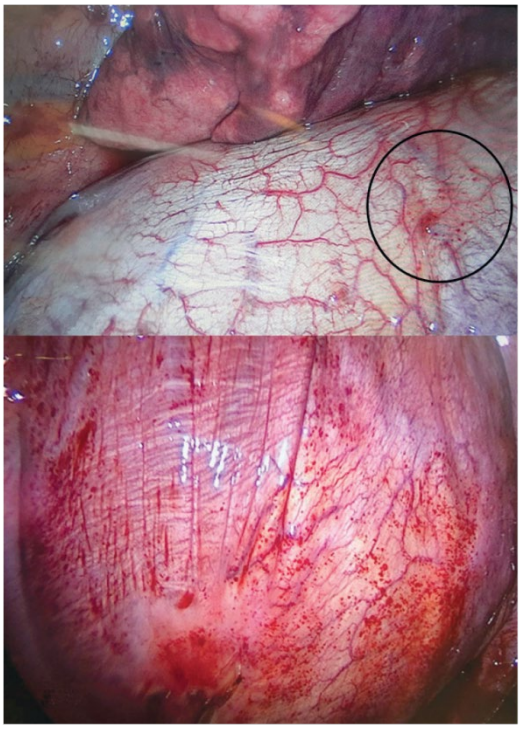

Fig. 4. Petechial punctate rashes on the central tendon of the diaphragm. A — circled; B — over the entire surface of the central tendon of the diaphragm

Lung resection (atypical, marginal) was performed in 10 patients (38.5%). Indications for lung resection were the presence of focal and bullous changes. It should be noted that 5 samples out of 8 were taken from the apical segment of the upper lobe, and in 3 patients - from the segments of the middle and lower lobes. Nevertheless, histological examinations of 8 out of 10 resected lung sections showed the presence of emphysematous changes in the lung. However, none of the women had any bad habits or working conditions associated with smoke, dust, etc. According to the literature, changes characteristic of bullous disease were detected in 81.3% of women with ERP and CP during resection of the apical part of the lung [29]. These changes are highly likely to be the result of natural physiological processes described in textbooks on respiratory physiology [30]. In none of our observations were foci of endometriosis detected in lung samples taken from women with CP. According to literature data, visceral pleural endometriosis was observed in 49.4% of patients. According to Japanese authors, endometrioid stroma with a positive reaction to receptors for estrogen, progesterone and CD 10 was detected in the walls of apical blebs in CP and ERP, which significantly expanded our understanding of the polymorphism of endometrioid heterotopias [21].

We consider the quality and reliability of pleurodesis to be a fundamentally important issue in the treatment for CP. Early recurrence of pneumothorax, recorded in the period after surgical treatment and before discharge, was observed in 12 patients (46.1%), the median day after surgery was 6 (3; 6.75) with a maximum value of 9 days. Two patients had persistent air leakage through the drainage tubes lasting for 14 days. In one patient, the source of air leakage was the suture line of one resected lung segment; in the second patient, lung resection was not performed, and the source was not verified. In 5 of 10 women, early recurrence of pneumothorax was clearly related to mensis and was also catamenial. In 2 of the 5 patients (3 and 7 days after surgery), plain chest radiography revealed free abdominal gas, which was not present on previous radiographs (Fig. 5). Following abdominal gas, chest radiographs showed evidence of localized recurrent pneumohydrothorax. These patients underwent chest drainage. Two of them had additional pleurodesis with doxycycline via a chest tube, and one patient was discharged with a chest tube and Heimlich valve. One patient with persistent air leakage after doxycycline pleurodesis developed a localized pleural empyema that was successfully treated with drainage without removal of the mesh implant.

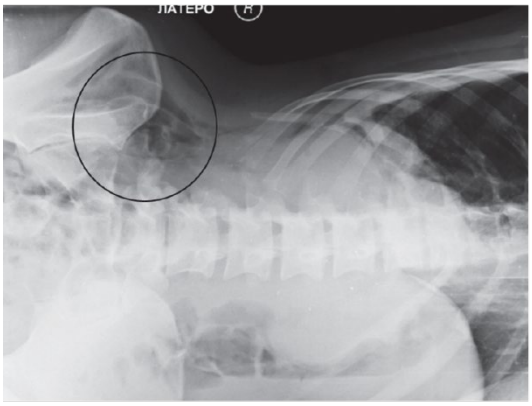

Fig. 5. Direct laterogram with the right side up. Free gas in the abdominal cavity (circled) on the 7th day after the operation, coinciding with the onset of mensis

Hormonal therapy prescribed by the obstetrician-gynecologist included the administration of the drug on the 2nd day of the first menstruation after surgery. If the time of drug administration was 1-2 weeks after surgery, it did not provide suppression of menstrual function, since the effect of the drug is achieved only by the 21st day after its intramuscular administration. Thus, the early postoperative period, when pleurodesis should be formed, was not protected from CP recurrence.

What problematic aspects should be highlighted from more than 10 years of experience in treating patients with this pathology? Catamenial pneumothorax has 2 pathogenetic variants of development. The first is when there is a predominantly genital-abdominal-transdiaphragmatic route of air entry into the pleural cavity. This type of pneumothorax is rarely large in volume and is sometimes an accidental finding of the radiologist. The second is when CP occurs due to the destruction of lung tissue at the site of implantation of endometrioid heterotopias under the visceral pleura, which can happen both with visible fenestrations of the diaphragm and without them. Probably, it is precisely with damage to the visceral pleura that pneumothorax occurs, accompanied by chest pain, shortness of breath, that is, it is a reason for emergency surgery. In the first variant and small-volume pneumothorax, we often do not drain the pleural cavity, but limit ourselves to dynamic observation of patients, and have time and opportunity for a full gynecological examination and identification of the recurrent nature of pneumothorax. Then, with a combination of anamnesis and radiography data, we establish a diagnosis of recurrent pleurodesis. In this situation, when planning the operation, it is advisable to preemptively administer hormonal therapy with drugs from the GnRH agonist group 1–2 months before the intervention in order to suppress menstruation and protect "immature" pleurodesis from recurrent pneumothorax with subsequent continuation of complex course therapy after surgery. This excludes the development of early relapses in the postoperative period. A similar strategy was proposed by M.B. Marshall in 2005 [4]. Considering the possibility of combination of both variants, it is necessary to carefully revise the lung and remove all suspicious areas by the method of economical marginal resection, both for the prevention of pneumothorax recurrence and for histological analysis and IHC examination. After performing a surgical intervention without preventive hormonal therapy, the timing of the next menstruation after surgery should be taken into account, without removing the drainage tubes before this time. However, such tactics do not guarantee complete protection against relapse, since relapses are more often local. In case of recurrence of catamenial pneumothorax, drainage of the pneumothorax cavity and additional chemical pleurodesis through a drainage tube are indicated. This intervention was performed in 4 patients within about 1 month after surgery.

The most difficult issue is the diagnosis of visceral pleural endometriosis. On the one hand, when removing lung areas with a bleb or thin-walled subpleural bulla up to 2 cm in diameter, no endometrioid heterotopias were detected in any observation. On the other hand, marginal lung resection increases the risk of prolonged air leakage through the drains, which is confirmed by foreign literature data [31; 28], and this can also reduce the quality of pleurodesis. The solution to this problem in light of the study of the long-term outcomes of various methods of surgical treatment and the use of hormonal therapy will be the next stage of research into this problem.

CONCLUSION

When detecting pneumothorax, especially right-sided, in women of childbearing age, it is necessary to collect anamnesis in order to identify clinical manifestations of both internal and thoracic

endometriosis; to determine the phase of the menstrual cycle at the time of occurrence of clinical manifestations of the disease; to recommend a full gynecological examination and performing an X-ray examination of the lungs on the 1st-2nd day of mensis during two or three menstrual cycles. If recurrent catamenial pneumothorax is confirmed, further complex treatment and subsequent dynamic observation by a multidisciplinary team consisting of the thoracic surgeon and obstetrician-gynecologist are indicated. Together with the pathologist, it is necessary to optimize the diagnosis of endometriosis using immunohistochemistry. The surgery should provide a full revision and removal of all obvious and questionable emphesematous formations and macroscopic foci of the lung, diaphragm and parietal pleura. Since it is not possible to ensure complete removal of all microscopic endometrioid heterotopias of the diaphragm within the scope of the operation, then, in order to improve the immediate outcomes of surgical treatment of catamenial pneumothorax, we see the need for preventive administration of hormonal therapy 1.5–2 months before the planned surgical intervention, followed by continuation of therapy in the postoperative period.

REFERENCES

1. *Endometrioz* – 2020 (02.11.2020). Klinicheskie rekomendatsii. Moscow, 2020. Available at: http://disuria.ru/_ld/10/1002_kr20N80mz.pdf?ysclid=lmt188qxlh336010287 [Accessed Sep 21, 2023]
2. Rousset-Jablonski C, Alifano M, Plu-Bureau G, Camilleri-Broet S, Rousset P, Regnard JF, et al. Catamenial pneumothorax and endometriosis-related pneumothorax: clinical features and risk factors. *Hum Reprod*. 2011;26(9):2322–2329. PMID: 21685141 https://doi.org/10.1093/humrep/der189
3. Joseph J, Sahn SA. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. *Am J Med.* 1996;100(2):164–170. PMID: 8629650 https://doi.org/10.1016/s0002-9343(97)89454-5
4. Marshall MB, Ahmed Z, Kucharczuk JC, Kaiser LR, Shrager JB. Catamenial pneumothorax: optimal hormonal and surgical management. *Ann R Coll Surg Engl*. 2005;27(4):662–666. PMID: 17626909 https://doi.org/10.1016/j.ejcts.2004.12.047 PMID: 15784370
5. Alifano M, Jablonski C, Kadiri H, Falcoz P, Gompel A, Camilleri-Broet S, et al. Catamenial and noncatamenial, endometriosis-related or nonendometriosis-related pneumothorax referred for surgery. *Am J Respir Crit Care Med*. 2007;176(10):1048–1053. https://doi.org/10.1164/rccm.200704-587OC
6. Ciriaco P, Negri G, Libretti L, Carretta A, Melloni G, Casiraghi M, et al. Surgical treatment of catamenial pneumothorax: a single centre experience. *Interact Cardiovasc Thorac Surg*. 2009;8(3):349–352. PMID: 19088096 https://doi.org/10.1510/icvts.2008.190975
7. Korom S, Canyurt H, Missbach A, Schneiter D, Kurrer MO, Haller U, et al. Catamenial pneumothorax revisited: clinical approach and systematic review of the literature. *J Thorac Cardiovasc Surg*. 2004;128(4):502–508. PMID: 15457149 https://doi.org/10.1016/j.jtcvs.2004.04.039
8. Voskresensky OV, Smoliar AN, Damirov MM, Galankina IE, Zhelev IG. Thoracic endometriosis and catamenial pneumothorax. *Pirogov Russian Journal of Surgery*. 2014;(10):4–9. (In Russ.)
9. Voskresensky OV, Damirov MM, Tarabrin EA, Galankina IE, Sharifullin FA. Catamenial (menstrual) pneumothorax is a complication of extragenital endometriosis. *Ginecology*. 2015;17(2):36–40. (In Russ.)
10. Hallifax RJ, Yousuf A, Jones HE, Corcoran JP, Psallidas I, Rahman NM. Effectiveness of chemical pleurodesis in spontaneous pneumothorax recurrence prevention: a systematic review. *Thorax*. 2017;72(12):1121–1131. PMID: 27803156 https://doi.org/10.1136/thoraxjnl-2015-207967
11. Alfageme I, Moreno L, Huertas C, Vargas A, Hernandez J, Beiztegui A. Spontaneous Pneumothorax: Long-term Results with Tetracycline Pleurodesis. *Chest*. 1994;106(2):347–350. PMID: 7774300 https://doi.org/10.1378/chest.106.2.347
12. Haga T, Kurihara M, Kataoka H, Ebana H. Clinical-pathological findings of catamenial pneumothorax: comparison between recurrent cases and non-recurrent cases. *Ann Thorac Cardiovasc*. 2014;20(3):202–206. PMID: 23698374 https://doi.org/10.5761/atcs.oa.12.02227
13. Haga T, Kataoka H, Ebana H, Otsuji M, Seyama K, Tatsumi K, et al. Thoracic endometriosis-related pneumothorax distinguished from primary spontaneous pneumothorax in females. *Lung*. 2014;192(4):583–587. PMID: 24831784 https://doi.org/10.1007/s00408-014-9598-1
14. Ziedalski TM, Sankaranarayanan V, Chitkara RK. Thoracic endometriosis: a case report and literature review. *J Thorac Cardiovasc Surg*. 2004;127(5):1513-1514. PMID: 15116017 https://doi.org/10.1016/j.jtcvs.2003.12.016
15. Fukuoka M, Kurihara M, Haga T, Ebana H, Kataoka H, Mizobuchi T, et al. Clinical characteristics of catamenial and non-catamenial thoracic endometriosis-related pneumothorax. *Respirology*. 2015;20(8):1272–1276. PMID: 26306694 https://doi.org/10.1111/resp.12610
16. Ochi T, Kurihara M, Tsuboshima K, Nonaka Y, Kumasaka T. Dynamics of thoracic endometriosis in the pleural cavity. *PLoS One*. 2022;17(5):e0268299. PMID: 35544515 https://doi.org/10.1371/journal.pone.0268299 eCollection 2022.
17. Pishchik VG, Obornev AD, Atyukov MA, Petrov AS, Kovalenko AI. Experience of treatment of endometriosisrelated pneumothorax. *Grekov's Bulletin of Surgery*. 2017;176(3):56–60. (In Russ.) https://doi.org/10.24884/0042-4625-2017-176-3-56-60
18. Pechenikova VA, Kostyuchek DF. Extragenital Endometriosis: Clinicopathologic and Immunohistochemical Analysis of 45 Observations With Diverse Organ Localization. *Journal of Obstetrics and Women's Diseases*. 2010;51(2):69–77. (In Russ.)
19. Bagan P, Le Pimpec Barthes F, Assouad J, Souilamas R, Riquet M. Catamenial pneumothorax: retrospective study of surgical treatment. *Ann Thorac Surg*. 2003;75(2):378–381. PMID: 12607643 https://doi.org/10.1016/s0003-4975(02)04320-5
20. Visouli AN, Zarogoulidis K, Kougioumtzi I, Huang H, Li Q, Dryllis G, et al. Catamenial pneumothorax. *J Thorac Dis*. 2014;6(Suppl 4):S448–S460. PMID: 25337402 https://doi.org/10.3978/j.issn.2072-1439.2014.08.49
21. Yukumi S, Suzuki H, Morimoto M, Shigematsu H, Sugimoto R, Sakao N, et al. Two Young Women with Left-sided Pneumothorax Due to Thoracic Endometriosis. *Intern Med*. 2016;55(23):3491–3493. https://doi.org/10.2169/internalmedicine.55.7187 PMID: 27904115
22. Nezhat C, Lindheim SR, Backhus L, Vu M, Vang N, Nezhat A, et al. Thoracic Endometriosis Syndrome: A Review of Diagnosis and Management. *JSLS*. 2019;23(3):e2019.00029. PMID: 31427853 https://doi.org/10.4293/JSLS.2019.00029

23. Bobbio A, Canny E, Mansuet Lupo A, Lococo F, Legras A, Magdeleinat P, et al. Thoracic Endometriosis Syndrome Other Than Pneumothorax: Clinical and Pathological Findings. *Ann Thorac Surg*. 2017;104(6):1865–1871. PMID: 29054304 https://doi.org/10.1016/j.athoracsur.2017.06.049
24. Alifano M, Roth T, Broët SC, Schussler O, Magdeleinat P, Regnard JF. Catamenial pneumothorax: a prospective study. *Chest*. 2003;124(3):1004–1008. PMID: 12970030 https://doi.org/10.1378/chest.124.3.1004
25. Leong A.C., Coonar A.S, Lang-Lazdunski L. Catamenial pneumothorax: surgical repair of the diaphragm and hormone treatment. *Ann R Coll Surg Engl.* 2006;88:547–549. https://doi.org/10.1308/003588406X130732
26. Cieslik L, Haider SS, Fisal L, Rahman JAA, Sachithanandan A. Minimally Invasive Thoracoscopic Mesh Repair of Diaphragmatic Fenestrations for Catamenial Pneumothorax Due to Likely Thoracic Endometriosis: A Case Report. *Med J Malaysi*. 2013;68(4):366-7.
27. Gavrilov SG, Son DA, Churikov DA, Efremova OI, Shuljak GD. Ultrasonic geometry of synthetic endoprostheses after transabdominal preperitoneal hernioplasty of inguinal hernias. *Pirogov Russian Journal of Surgery*. 2020;(1):53–60. (In Russ.). https://doi.org/10.17116/hirurgia202001153
28. Cai H, Mao R, Zhou Y. Video-assisted subtotal parietal pleurectomy: an effective procedure for recurrent refractory pneumothorax. *BMC Surgery*. 2022;22(1):204–209. PMID: 35619102 https://doi.org/10.1186/s12893-022-01653-5
29. Alifano M, Legras A, Rousset-Jablonski C, Bobbio A, Magdeleinat P, Damotte D, et al. Pneumothorax recurrence after surgery in women: clinicopathologic characteristics and management. *Ann Thorac Surg*. 2011;92(1):322–326. PMID: 21718864 https://doi.org/10.1016/j.athoracsur.2011.03.083
30. West JB. *Respiratory Physiology—The Essentials*. 3rd ed. Williams & Wilkins, 1985. (Russ. Ed.: Uest Dzh. *Fiziologiya dykhaniya. Osnovy*. Moscow: Mir Publ.; 1988)
31. Dżeljilji A, Karuś K, Kierach A, Kazanecka B, Rokicki W, Tomkowski W. Efficacy and safety of pleurectomy and wedge resection versus simple pleurectomy in patients with primary spontaneous pneumothorax. *J Thorac Dis*. 2019;11(12):5502–5508. PMID: 32030269 https://doi.org/10.21037/jtd.2019.11.28

Received on 01/11/2023
Review completed on 15/01/2024
Accepted on 05/06/2024