

Case Report

Pleural Endometriosis – Case Report

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- Thoracic endometriosis syndrome
- Videothoracoscopy

Abstract

Objective: To report a rare case of pleural endometriosis with recurrent spontaneous pneumothorax

Case report: A case of pleural diagnosis was diagnosed retrospectively at 51 years old. Expectation treatment was performed, including hormonal therapy and apical lobectomy with pleurodesis.

Conclusion: It is suggested that pleural endometriosis is a complex disease causing numerous diagnostic problems among clinicians. That is why majority of these cases are under diagnosed.

INTRODUCTION

Endometriosis affects between 5% - 15% of women in reproductive age, whereas no more than 12% of patients have ectopic tissue outside lower pelvis [1,2]. Minute fragments of active endometrial tissue may disseminate in distant areas of the peritoneal cavity, the umbilical region, lymph nodes, lungs, bones and heart. Endometriosis is a frequent cause of infertility, painful periods and pelvic pain. The first case of pleural endometriosis was described by Schwarz in 1938 [3]. TES is a constellation of four clinical presentations, catamenial pneumothorax, hemoptysis, hemothorax and pulmonary nodules [4]. TES may present with catamenial pneumothorax which occurs within 72 hours before or after menstruation [1,5]. The first case of catamenial pneumothorax was described in 1958 by Mauer et al. [6], and the term "catamenial pneumothorax" was first introduced into the literature in 1972 by Lillington et al. [7].

CASE PRESENTATION

51-year-old patient (E.W.) presented to her GP for routine health check. She complained of decreased exercise tolerance and effort dyspnea. Physical examination did not show any abnormalities. She had a history numerous admissions to pulmonology wards and menstrual problems (copious and painful menstruations). She had regular periods and was not treated for any chronic condition. At the age of 43 she had an emergency admission to pulmonology ward because of weakness, dyspnea and right-sided chest pain caused by spontaneous pneumothorax, diagnosed 5 days earlier on routine plain chest X-ray.

On admission, she presented with decreased oxygen saturation - SpO₂ 91% and tachypnea – 22 per minute. Laboratory

tests and ECG did not show any abnormalities. A series of chest x-ray films in antero and lateral projection revealed 5 mm air deposit in the right apical region. The 3 consecutive imaging performed every 2 days showed regression of the pneumothorax. Computerized tomography (CT) of chest without contrast performed on the day 7 did not show presence of any air in the right pleural cavity, no emphysematous bullae and no focal lesions in the lung parenchyma. Pleural recesses and diaphragm were normal. Ultrasound of abdominal cavity and thyroid were normal. The treatment included oxygen therapy (10 L/min) and estazolam. Patient was discharged home in good general condition and referred to outpatient chest clinic for follow-up.

Next month, the patient was re-admitted as an emergency to the thoracic surgery ward because of dyspnea, weakness and pain in the region of the right apex. Standard laboratory tests and ECG results were normal. X-ray examination revealed recurrent spontaneous right-sided pneumothorax (Figure 1). Right pleural cavity drainage was performed through the 4th intercostal space in the anterior axillary line. The drain was removed after obtaining clinical and radiological improvement. Pharmacotherapy included anti-coagulation, pain control (NSAIDs and opioids) and antibiotics (2nd generation cephalosporin). Patient was discharged home in good general condition and referred to outpatient chest clinic for follow-up.

One month later, the patient was again urgently admitted to the chest ward because of shortness of breath, weakness, and chest pain within the right apical region. Standard blood test and urine tests and ECG did not show any abnormalities. Chest x-ray showed recurrent spontaneous pneumothorax. It was treated conservatively with benzodiazepines. Control chest X-ray

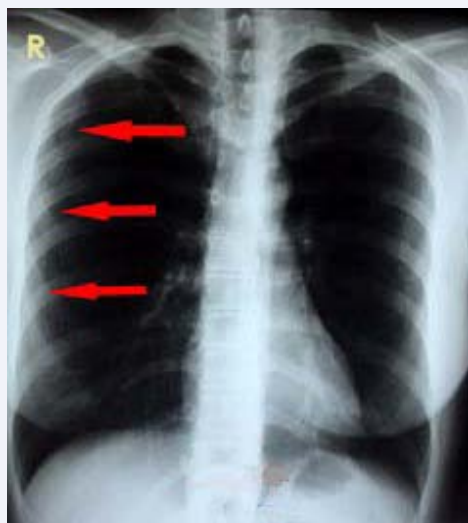


Figure 1 X-ray photo of right-sided pneumothorax, about 2cm.

showed spontaneous re-absorption. The patient was discharged in good general condition and referred to out-patient chest clinic for follow up.

Because of abundant and painful menstruation and suspicion of endometriosis on abdominal ultrasound examination, the patient was admitted to gynecology ward. Exploratory laparoscopy confirmed foci of endometriosis in the left ovary. During the procedure, a 15 mm biopsy sample of the left ovary was obtained and fallopian tubes were ligated subject to patient's consent. The histopathological diagnosis was: *Endometriosis externa*.

The patient was discharged in good general condition, referred for follow up in the outpatient gynae clinic and prescribed triptorelin as a continuation of treatment of endometriosis. Control chest X-ray performed during the treatment did not show any abnormalities. The treatment with triptorelin was discontinued after 1 month because of adverse effects - heat waves, vaginal dryness, head and breast pain, nausea and weakness. After interruption of the treatment, respiratory tract symptoms with recurrent pneumothorax recurred. A few months later, the patient was admitted to the thoracic surgery ward, where, following the video-thoracoscopy and pleuradesis, the patient was qualified for apical resection of the right lung. Three samples of lung tissue and pleura showed pulmonary and nodal pneumoconiosis, pleural thickening and fibrosis, focal emphysematous changes and congestion, subpleural fibrosis with eosinophilic infiltrate (Figure 2).

Histopathological slides and paraffin blocks were reviewed in Histopathological Diagnostic Laboratory of the Department of Pathology of the Medical University of Silesia. Examinations of routine serial H&E stained sections and immunohistochemical studies for the presence of estrogen receptor (ER) were performed. Apart from the changes reported in the original examination, also clusters of cells with positive ER reaction were found deep the pleura (Figure 3). A diagnosis of pulmonary endometriosis (*Endometriosis pulmonum*) was given. At present, the patient is under observation of chest physician, GP and gynecologist,

who implemented combined contraception. The patient has no respiratory tract symptoms as well any abnormalities on imaging.

DISCUSSION

Distant complications of cesarean section include increased risk of endometriosis. Therefore, constantly growing number of cesarean sections (an increase from 26% to 36, 5% in years 2003-2009 in the USA [8]) will be the main factor of increasing incidence of pelvic endometriosis as well as distant endometriosis, e.g. pleural and chest.

Population studies by Haga et al., enabled development of point system risk assessment of pleural endometriosis. Predictive factors include: right sided pneumothorax, pelvic endometriosis, age above 31 years, non-smoking (sensitivity 93.5% and specificity 89.4% with the result of 12 points or more) [1]. In our case, the patient had 18/18 Hagascore points, which puts her into a high risk group.

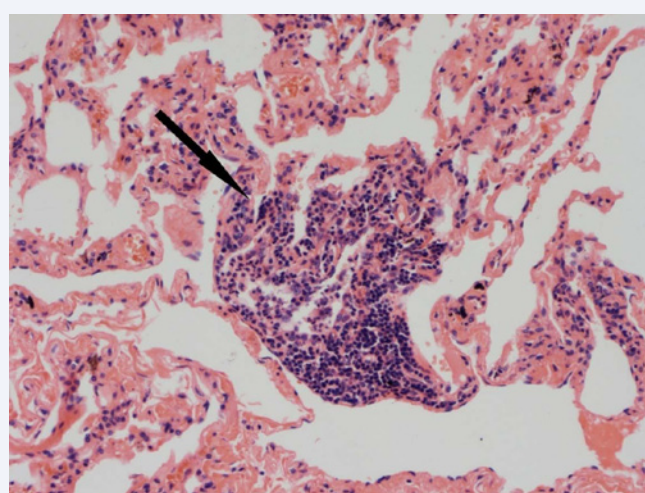


Figure 2 Areas of congestion of the lungs, inflammatory foci of eosinophils (→). Features of emphysema (100x zoom).

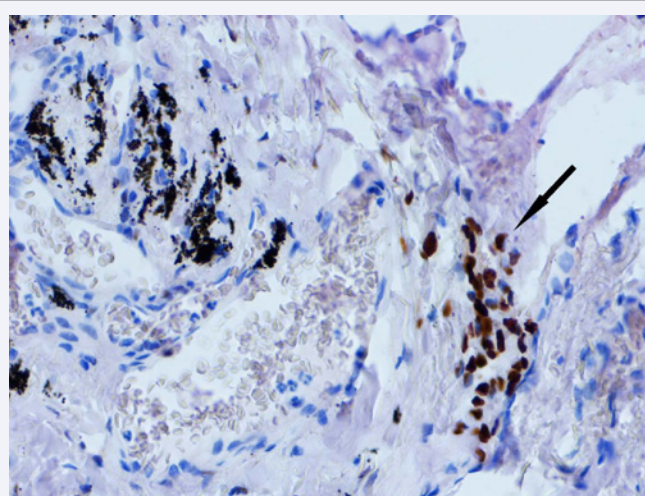


Figure 3 Staining for nuclear estrogen receptor. ER (+) (→) (200x zoom).

In the majority of cases, pleural endometriosis is accompanied by chest discomfort and pain, which sometimes may be neglected by patients [9]. Our patient confirmed the early onset of respiratory tract symptoms but of lower intensity. Numerous hospital admissions to pulmonology and thoracic surgery wards and diagnoses emphasize repetitiveness and cyclic nature. As discovered retrospectively, dyspnea preceded admissions, which supports the diagnosis of catamenial pneumothorax.

Numerous epidemiological studies indicate cause-effect relationship between cesarean section and the incidence of endometriosis [3]. Therefore, it is not surprising that ectopic foci of endometrial tissue were found. Fallopian tube ligation is believed to be preventive of further dissemination of endometrial tissue via fallopian tube, and then through the fenestrations in the diaphragm into the chest [10].

Patient's induction into artificial menopause was correlated with regression of respiratory tract symptoms and arrest of pneumothorax recurrence. This observation is an indirect indication of the presence of pulmonary endometriosis, which should be taken into account in the differential diagnosis. One should emphasize that restoration of natural menstrual cycle resulted in recurrence of dyspnea and pneumothorax. It was caused by activation of ectopic endometrium within the lungs.

As in a case presented by Zeena Makhija et al., videothoracoscopy (gold standard in diagnosis and treatment of pleural endometriosis) was performed with lobectomy and pleuradhesion (creation of adhesions between visceral and parietal pleura) [11]. The procedure brought a satisfactory therapeutic effect. VATS procedure included close inspection of chest cavity, including diaphragmatic aspect, which did not show any abnormalities. Presence of visible fenestrations in diaphragm may explain the pathway of endometrial tissue as it was in case described above [11]. However, invisible micro-fenestrations may also be seen sites for the ectopic tissue [3]. The hypothesis about hemato- and/or lymphogenic dissemination is also possible considering the fact of surgical intervention within the uterus (cesarean section) in the past [7]. Another hypothesis regarding the etiology of endometriosis relies on the possibility of metaplasia body cavity lining into endometrial cells [7].

The ultimate diagnosis and confirmation is based on the histopathological examination. H&E staining usually does not allow detecting foci of endometriosis. The recommended method to detect ectopic endometrial tissue is immunohistochemistry (cytokeratin-7, BER-EP4, and estrogen and progesterone receptors) [12]. A characteristic finding was presence of eosinophilic inflammatory infiltrate and numerous macrophages. Similar histopathological results were obtained in case reported by Yoshinobu Ichiki et al. [9]. In a case of pleural endometriosis reported by Mesbah Rahman, routine H & E stain did not reveal endometrial structures but additional immunohistochemical examinations detected endometrial cells [13].

TES is a complex disease causing numerous diagnostic problems among clinicians as well as among pathologists. Spontaneous recurrent pneumothorax that accompanies menstruation in women with diagnosed pelvic endometriosis [2,14]. Important findings that may help to establish the diagnosis is presence of small or large endometrial foci, undergoing hormonal influence, seen during surgical procedures. Diagnosis of TES requires experience and knowledge. The diagnosis and appropriate therapeutic approach are especially important for successful treatment.

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