



Pulmonary complications requiring surgical intervention caused by endometriosis – case studies

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Abstract

Endometriosis is a condition described as growth of the endometrium outside the uterine cavity. Lesions can occur in many areas of the body, including the pleural cavity and lungs. Etiology of this condition is still unknown. Two medical cases are described: a 47-year-old patient reporting chest pain, who had resection of the apex of the right lung a year earlier, and 41-year-old patient with frequent lower abdominal pain and recurrent haemoptysis for about 16 years, which had been the cause of multiple hospitalizations in pulmonary departments. Both patients underwent several examinations – tomography, MRI, and surgical diagnostics. In both cases, the tests showed the presence of thoracic endometriosis. The described cases indicate diagnostic and therapeutic difficulties in patients with thoracic endometriosis. In conclusion, treatment of the manifestation of thoracic endometriosis is a great challenge because of limitation of the possibilities for surgical procedures and non-invasive diagnostic tests.

Key words

endometriosis, thoracic endometriosis, catamenial pneumothorax, catamenial haemoptysis

Abbreviations

CT – computed tomography; **VATS** – video-assisted thoracoscopic surgery; **GnRH** – gonadotropin-releasing hormone; **TES** – thoracic endometriosis syndrome; **TVUS** – transvaginal ultrasound

INTRODUCTION

Endometriosis is described as growth of the endometrium outside the uterine cavity and is associated with characteristic symptoms, such as dysmenorrhea, menorrhagia, and chronic pelvic pain, and causes infertility [1]. The disease affects 15% among reproductive-age women [2, 3]. Endometriosis has a strong impact on psychological and social well-being, causing depression, sexual dysfunction and inability to work. Treatment also generates significant costs (long diagnosis, surgical therapy, hospital admission, fertility therapy) [4].

Laparoscopy is the gold standard in the diagnostic process. One of the main reasons for the delay of a final diagnosis of this condition is because it requires surgery and histological examination of the tissue [4,5]. Endometrial lesions may present in many places in the pelvis, including the uterus (adenomyosis), ovary (endometrioma), pelvic peritoneum, bladder/ureter, rectum, colon, uterosacral ligaments, rectovaginal septum, vaginal wall, or pouch of Douglas. Endometrial implants may also occur at many rare locations in the body, such as the lungs, liver, pancreas, brain and C-section scar, resulting in a variety of symptoms related to these organs [4]. The most common location of endometriosis outside the abdominopelvic cavity is the thoracic cavity [6]. In 80% of cases, lesions are located on the right side [7, 8].

The presence of functional endometrial tissue in the thoracic cavity is called thoracic endometriosis syndrome (TES), an extremely rare disease with complex causes and pathogenesis that remains unclear [7, 9], and can be classified as pulmonary or pleural [8]. The pathogenesis of this disease is still unknown although several concepts have been presented, but none of them can fully explain all the clinical manifestations of the syndrome [9]. Retrograde menstruation is the most prominent theory, which assumes that endometrial cells undergo a retrograde movement through the fallopian tubes into the peritoneal cavity, and implant on peritoneal surfaces [6, 10]. The coelomic metaplasia theory assumes that endometriosis is formed by metaplasia of mesothelial cells lining the pleura and peritoneal surfaces into endometrial glands and stroma. Transformation of these cells may be stimulated by estrogens [6]. Although the coelomic metaplasia theory may provide an explanation for pleural cases of endometriosis, the bronchopulmonary lesions remain unexplained [6, 8].

On the other hand, a possible explanation for bronchopulmonary endometrial lesions is the theory of benign metastases, which proposes that ectopic endometrial implants are the result of lymphatic or haematogenous dissemination of endometrial cells [2, 6]. The final approach explaining TES involves prostaglandin F_{2α} which is detectable in the plasma of women during menstruation. Prostaglandin F_{2α} is a constrictor of bronchioles and blood vessels, which increases during menstruation and may lead to alveolar rupture of previously formed subpleural blebs and bullae, resulting in a pattern of catamenial pneumothorax [7, 10].

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Pelvic endometriosis expressions typically occur approximately 5–7 years before developing manifestations of thoracic endometriosis [6]. Symptoms of pulmonary endometriosis are always associated with the onset of menses and usually manifests as catamenial haemoptysis [8,11]. Haemoptysis in the majority of cases stop after the cessation of menstruation and may be accompanied by chronic cough, catamenial pneumothorax, episodes of low grade fever which may be recurrent, and asymptomatic lung nodules [3].

OBJECTIVE

The presented case reports aim to demonstrate the diagnostic and therapeutic difficulties in patients with thoracic endometriosis, and also to indicate the difficulties in conducting therapy and the need for a multidisciplinary and individual approach to the patient.

MATERIALS AND METHOD

The material for this case study was collected from the real-life clinical process and medical records of the gynaecological ward at the Women's and Child Health Centre in Zabrze, Poland.

In accordance with the Helsinki Declaration, the case reports were fully anonymised, and none of the data presented would make identification of the patients possible. Under Polish law, such case reporting does not require the consent of a Bioethics Committee.

CASE STUDIES

Patient 1

A 47-year-old patient treated chronically for endometriosis reported to the attending physician complaining of pain in the right side of the chest. Due to the ailments described by the patient, a chest X-ray and a CT chest scan were performed. In the right pleural cavity, fluid, atelectasis and fibrous changes were visualized. At the time of reporting to the attending physician, the patient complained of a stinging pain in the chest while breathing and performing movements, and reduced exercise tolerance. The attending physician ordered a pelvic MR examination which showed the features of intensified deeply infiltrating endometriosis, with ingrown lesions in the dorsal part of the sigmoid colon and the area of the large intestine at the border of the sigmoid colon and rectum. Surgical consultation was recommended.

The patient was referred to the thoracic surgery ward where right-sided recurrent pneumothorax with exudate to the pleural cavity was diagnosed. The pneumothorax was decompressed by drainage of the pleural cavity. Past medical history included resection of the apex of the right lung a year earlier. The material collected during the operation confirmed endometriosis of the lungs and pleura. The attending physician initiated treatment with the Diphereline SR (Triptorelinum) in a dose of 3.75 mg with prolonged release administered intramuscularly. After the administration of six doses of Diphereline, treatment with Depo-Provera (medroxyprogesteroni acetat) was initiated by intramuscular injection every 90 days.

At the follow-up visit six months later, the patient did not report any pain in the pelvic area, nor the symptoms of pneumothorax described earlier. During the visit, a transvaginal pelvic ultrasound was performed which showed a tumour located behind the uterus that may represent an endometriosis tumour (Fig. 1). It was therefore decided to continue the supply of Depo-Provera. The treatment cycle was completed after the eighth intramuscular injection of Depo-Provera. The last medical appointment took place in 2021. The patient was in good general condition and did not report any complaints.

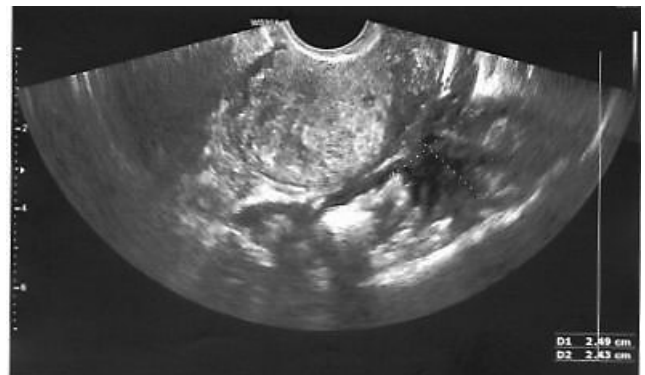


Figure 1. Patient 1. Transvaginal pelvic ultrasound, uterus, a tumour located behind the uterus that may represent an endometriosis tumour.

Patient 2

The 41-year-old patient had been hospitalized several times due to pain in the lower abdomen, and reported suffering periodic haemoptysis for about 16 years, which resulted in repeated hospitalization in pulmonary departments. Endometriosis was suspected and confirmed by laparoscopy in 2013. The patient described the pain in the lower abdomen as jerking, stinging, pulling, burning, rushing, but not related to the phase of the menstrual cycle. Pain and haemoptysis made it difficult to perform daily activities. The patient underwent multiple surgical consultations due to repeated episodes of severe lower abdominal pain with suspected acute abdomen.

Transvaginal pelvic ultrasound showed an adhesion and intrauterine endometrial lesions (Fig. 2, Fig. 3). Repeated haemoptysis was the basis for extending pulmonary diagnostics. In 2018, the decision was made to re-laparoscopy



Figure 2. Patient 2. Transvaginal pelvic ultrasound, ovary, adhesion

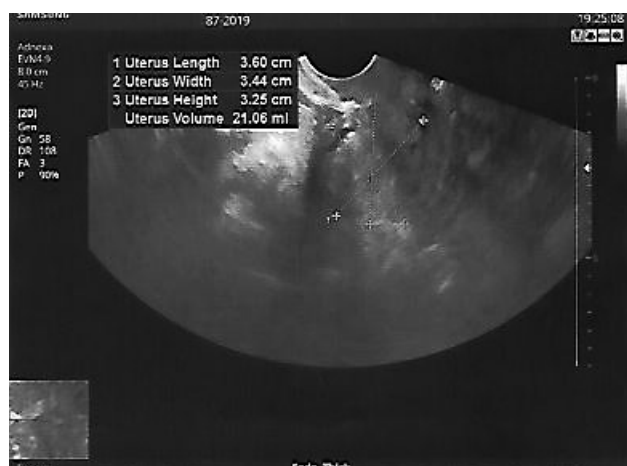


Figure 3. Patient 2. Transvaginal pelvic ultrasound, uterus, intrauterine endometrium

to release the adhesions, and perform electrocautery of the right ovary. Endometriosis foci in the small pelvis were coagulated. A CT of the chest was ordered which showed a lesion that could correspond to the endometriosis lesion in segment VI. Due to the suspicion of thoracic endometriosis, it was decided to perform interventional bronchoscopy. Due to persistent pain in the lower abdomen, haemoptysis and heavy breathing, treatment with Diphereline SR (Triptorelinum) in a dose of 3.75 mg was started for six months

Table 1.

| Case | Timeline | Event |
|-----------|-----------|--|
| Patient 1 | 1998 | Surgical resection of ovarian lesions. |
| | 2019 | VATS surgery - resection of the apex of the right lung, pleurectomy and pleurodesis. |
| | 2019 | Lesions in TVUS – suspicion of endometrial tumour. |
| | 2020 | Pneumothorax - histopathological examination confirmed thoracic endometriosis. |
| | 2022 | Lesions of pleura in CT – suspicion of pleural endometriosis. |
| | 2022 | Pneumothorax - hospitalization in the thoracic surgery department, decompression and drainage. |
| Patient 2 | 2004 | First symptoms: lower abdominal pain. |
| | 2013 | First laparoscopy due to endometriosis. |
| | 2014 | Endometrial cyst of the left ovary in TVUS. |
| | 2017–2019 | Multiple hospitalization for lower abdominal pain. |
| | 2018 | Second laparoscopy due to endometriosis. |
| | 2022 | Bronchial nodule in CT – suspicion of thoracic endometriosis. |
| | 2022 | Interventional bronchoscopy. |

DISCUSSION

The presented cases describe the diagnostic and therapeutic difficulties in patients with pulmonary endometriosis. The results were compared with currently available literature in which it was noticed that in the described cases difficulties were also experienced in diagnosing the disease. Bricelj et al. conducted a systematic review and reported that 39.5% of

patients with endometrial pneumothorax had coexisting pelvic endometriosis [12]. The same study reported that people diagnosed with pelvic endometriosis had more endometrial implants in the chest [12]. Despite the implementation of appropriate treatment and surgery, symptoms recurred in 26.9% of patients, [12]. Kardaman et al., in case report describe the clinical case of a 48-year-old female patient with recurrent pneumothorax on the right side. The authors describe that endometrial changes were more common on the right side, which was also found in *Patient 1* [13]. Researchers from the Pneumology Department, Farhat Hached Hospital in Sousse, Tunisia, described the case of a 42-year-old woman with recurrent right-sided pneumothorax. She underwent video-assisted thoracoscopic surgery (VATS), during which numerous endometrial lesions were visualized and confirmed by histopathological examination [11]. The patient was treated with a GnRH agonist, as was the case with *Patient 1*. Also in this case, no permanent remission was achieved, and the symptoms recurred after 22 months [11]. The case report by Leonardo-Pinto et al. also describes pulmonary manifestations of endometriosis [14] in the case of a 23-year-old female patient with haemoptysis occurring for two years around the time of menstruation. A nodule was found in the right lung that might correspond to a lesion of endometriosis; transvaginal ultrasound confirmed deeply infiltrating endometriosis of the reproductive organs. Surgical treatment was not performed in this patient. Based on the symptoms, empirical treatment was implemented, achieving an improvement in the clinical condition [14]. Researchers from the Department of Thoracic and Cardiovascular Surgery, Uijeongbu St. Mary's Hospital, The Catholic University of Korea in Seoul, Republic of Korea, describe surgical intervention as an effective means of preventing the recurrence of haemoptysis, based on the case report of a 23-year-old patient with catamenial haemoptysis [15]. After resection of the diseased lung fragment, no recurrence of clinical manifestations of the disease was observed during the 5-year follow-up. [15]. The current case reports confirms the systematic review reported by researchers from the Department of Obstetrics and Gynecology at McGill University in Montreal, Canada. Observations in the current cases concur – the most common pulmonary endometriosis occurs on the right side, and are also in agreement with the literature [16].

Limitations of the case studies. In the two case reports it may be that the patients had different lengths of medical history, as well being of different ages. Currently, two more patients with catamenial haemoptysis are being monitored at the Women's and Child Health Center in Zabrze; therefore, expansion is planned in which further studies will be undertaken to check whether the occurrence of pulmonary complications in patients with endometriosis can be predicted before clinical manifestations occur.

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