CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 33 (2017) 139–142

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

A case report of thoracic endometriosis – A rare cause of haemothorax

Lina Pankratjevaite[a], Diana Samiatina-Morkuniene[b,*]

[a] Department of Surgery, Medical Academy, Hospital of Lithuanian University of Health Sciences, Kaunas, Lithuania
[b] Department of Thoracic Surgery, Medical Academy, Hospital of Lithuanian University of Health Sciences, Kaunas, Lithuania

ARTICLE INFO

Article history:
Received 19 November 2016
Received in revised form 27 February 2017
Accepted 27 February 2017
Available online 4 March 2017

Keywords:
Thoracic endometriosis syndrome
Catamenial haemothorax
Pleural effusion
Case report

ABSTRACT

INTRODUCTION: The presence of endometrial tissue in airways, pleura and lung parenchyma is called thoracic endometriosis syndrome (TES). It is a rare pathology, and typically consists of catamenial pneumothorax, haemothorax, haemoptysis, and pulmonary nodules. We report a case of a 36-year-old woman with thoracic endometriosis causing catamenial haemothorax.
CONCLUSIONS: The diagnosis of thoracic endometriosis is complicated and often delayed. TES should be suspected in a reproductive age woman with exacerbating symptoms during the menstruation.
Treatment may be medical and surgical.

1. Introduction

Endometriosis is defined as the presence of normal endometrial mucosa implantation outside the uterine cavity [1,2]. It is diagnosed in women of reproductive age [3]. Endometriosis most commonly affects pelvic organs, but it can be found in extra-pelvic organs and tissues [4,5]. One of extra-pelvic endometriosis form is thoracic endometriosis (TE) [3]. It can affect airways, pleura and lung parenchyma [6,7]. The presence of endometrial tissue in or around the lung is called thoracic endometriosis syndrome (TES) [2]. Typically, it consists of catamenial pneumothorax, haemothorax, haemoptysis, and pulmonary nodules [2,8]. The majority of patients commonly have catamenial pneumothorax (73%), while catamenial haemothorax is present just in 14% of the cases [9]. Chest pain, dyspnoea, cough, haemoptysis, and scapular pain are the most common patients' complaints [4]. Symptoms usually occur between 1 day before and 2–3 days after the onset of menstruation [8]. Diagnosis of TE is difficult. Anamnesis [10], imaging studies [2,3,8], histopathological examination [3] play an important role in the diagnosis of TES. However they all have limitations. The same symptoms are accompanied with other pulmonary diseases [4], radiological abnormalities are transient and there are no specific diagnostic criteria [4], and only one third of the cases may have histopathological confirmation of endometriosis diagnosis [11].

TES is treated medically and surgically. When medical treatment fails, surgical resection of endometriosis damaged tissue is suggested [12].

In line with the SCARE criteria, we report a case of a 36-year-old woman with thoracic endometriosis causing catamenial haemothorax [13]. It is the first case of TES that has required urgent surgical treatment in our hospital in past fifteen years.

2. Case report

A 36-year-old female was admitted to the Emergency Room (ER) because of pain in the right side of the abdomen and chest, breathlessness and faintness lasting few hours. The patient denied any trauma. Her past medical history was significant for long lasting iron deficiency anaemia (she had few blood transfusions), infertility and two abdominal surgeries. Ten years ago she underwent myomectomy, and seven years ago she had diagnostic laparoscopy because of severe bleeding during the menses (pelvic endometriosis was diagnosed). During physical examination, her blood pressure was 90/60 mmHg, and heart rate 89 times per min. Her skin and visible mucous were pale. Blood laboratory findings revealed anaemia: haemoglobin- 56 g/l, red blood cells – 2.63×10^{12}/l. Chest x-ray showed pleural effusion on the right side (See Fig. 1). One litre of haemorrhagic fluid from pleural cavity was drained in the ER. Then, a computed tomography scan of the chest, abdomen and pelvis was performed. It demonstrated a small amount of air and heterogenic fluid in the right side of the chest after right pleural cavity drainage, and a small amount of fluid in the abdomen. No more changes in the chest were detected. However, heterogenic nodules in the uterus, and solid density masses in the lower part of the abdomen were seen.

Hypovolemia was corrected by the transfusion of four units of red blood cells and crystalloid liquids in the ER. After that, the patient was admitted to the Department of Thoracic surgery for further examination and treatment. The patient was haemodinam-

* Corresponding author at: Department of Thoracic Surgery, Hospital of Lithuanian University of Health Sciences, Eiveniu 2, LT-50009, Kaunas, Lithuania.
E-mail address: dsamiatina@yahoo.de (D. Samiatina-Morkuniene).

http://dx.doi.org/10.1016/j.ijscr.2017.02.052

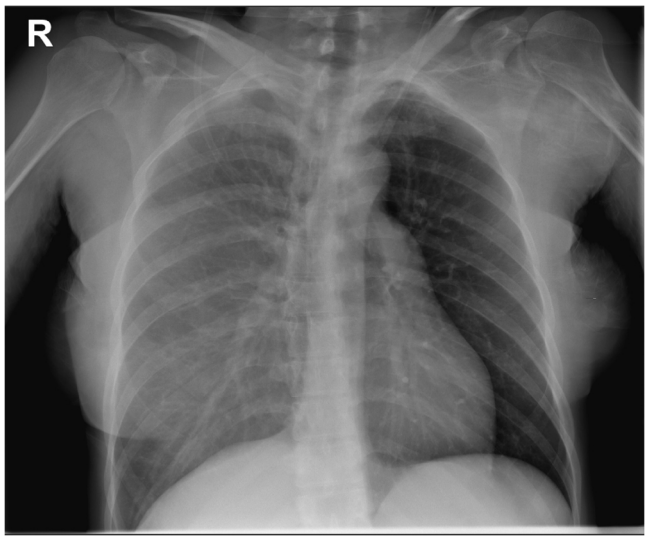

Fig. 1. Chest x-ray showed pleural effusion on the right side.

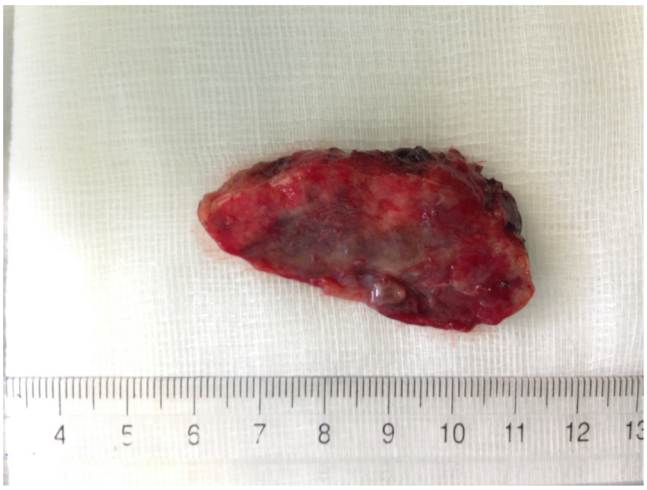

Fig. 3. Edometrial lesions, pulmonary nodules on the lung.

ically stable, the bleeding through the chest probe was stopped. Because of that, no urgent surgery was performed. Prior to video-assisted thoracoscopic surgery (VATS), an abdominal ultrasound was performed. It showed the same amount of fluid in the abdominal cavity. The patient underwent gynaecological examination. A transvaginal ultrasound examination revealed internal genitalia and pelvic endometriosis. However, severe bleeding through the chest probe repeated. The patient collapsed. It was the second day after the onset of menstruation. She was urgently taken to the operation theatre. Right side minithoracothomy was carried out. Blood clots in the right pleural cavity and suspected endometriosis lesions on the lungs, parietal pleura and diaphragm were found. The blood was slowly leaking from these lesions (See Fig. 2). Electrocoagulation, atypical lung resection was performed, and biopsy was taken (See Fig. 3). The lung was sutured with 3/0 Vicril suture. The pleurectomy of endometriosis affected pleura was also performed (See Fig. 4). Histopathological examination confirmed the diagnosis of endometriosis. The patient's postoperative course was uneventful. During the hospitalisation and three months after it, there was no recurrence of postoperative chest haemothorax. Due

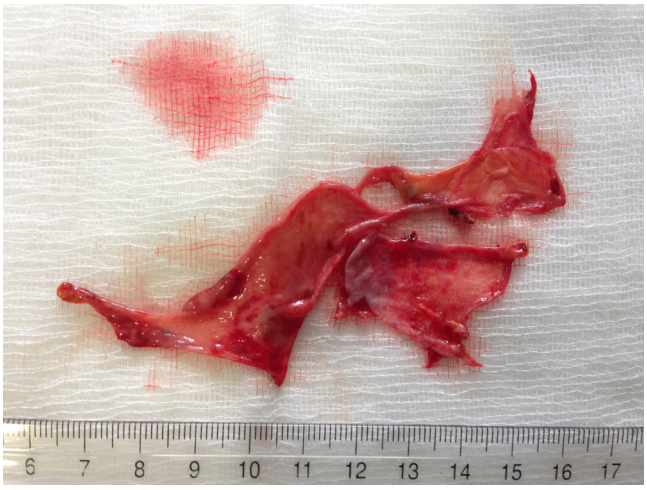

Fig. 4. Endometrial tissue affected pleura.

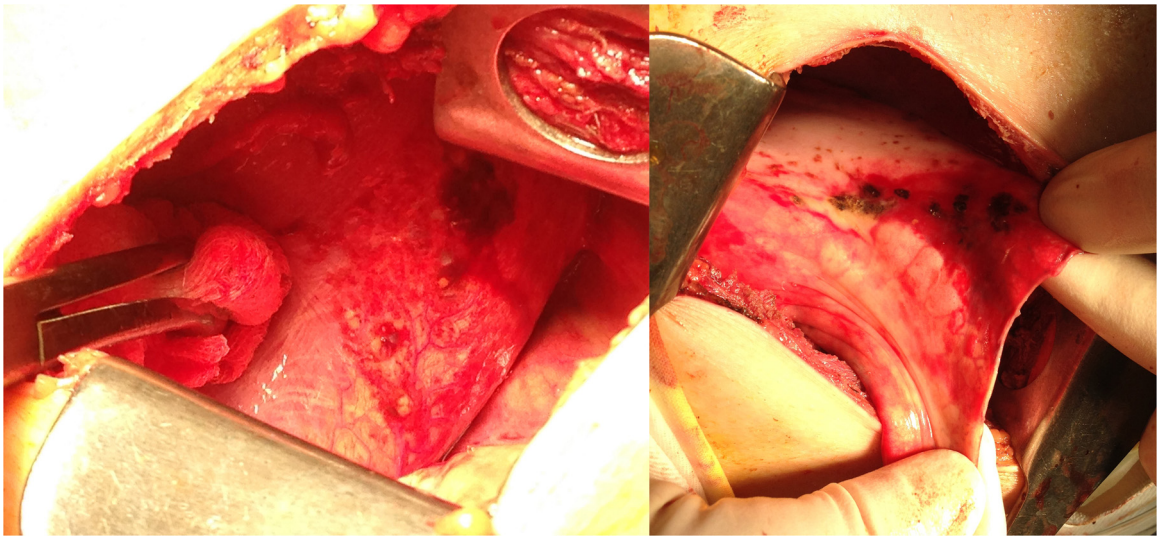

Fig. 2. The blood is slowly leaking from endometrial lesions.

to endometriosis, the patient was referred for further outpatient treatment.

3. Discussion

Endometriosis most commonly affects pelvic organs, but it can be found in extra-pelvic organs and tissues [4,5]. The presence of endometrial implants in airways, pleura and lung parenchyma is called thoracic endometriosis syndrome (TES) [2,6,7]. It consists of catamenial pneumothorax, catamenial haemothorax, catamenial haemoptysis, and pulmonary nodules [2,8].

The majority of patients commonly have catamenial pneumothorax (73%), while catamenial haemothorax is present just in 14% of the cases, followed by 7% of haemoptysis and 6% of lung nodules [9]. Moreover, TES is associated with pelvic endometriosis and infertility [6]. In our case, the patient suffered from the right side haemothorax. Her past medical history was significant for pelvic endometriosis and infertility. The lung nodules were found during surgery.

There are several different proposed theories for the formation of TE [4,7,8,10,14]. The first hypothesis is lymphatic or haematogenous embolization from the uterus [4,7,8,14,15], the second presents coelomic metaplasia theory [4,8,10,14,15], and the third proposes retrograde menstruation during which endometrial tissue from the uterus and fallopian tubes migrates via abdomen and congenital or acquired diaphragmatic defects into the pleural cavity [7,8,10,14,15].

Thoracic endometriosis syndrome is a rare pathology, and diagnosis is often delayed. Anamnesis [10], imaging studies [2,3,8], histopathological examination [3] play an important role in the diagnosis of TE. Chest pain, dyspnoea, cough, haemoptysis, and scapular pain are common patients' with TE complaints [4]. Symptoms usually occur between 1 day before and 2–3 days after the onset of menstruation [8], but they also can present in the intermenstrual period [4]. Moreover, the same symptoms may be accompanied by other pulmonary diseases such as pulmonary malignancy or tuberculosis [4]. During the physical examination, diminished or absent breath sounds on the affected side can be found [2]. Chest x-ray reveals normal [3] or non specific findings such as pneumothorax, pleural effusions, or pulmonary nodules [2]. Ultrasonography role is also important in diagnosis of endometriosis [1], due to the fact that TE may go together with pelvic and abdomen endometriosis. In our case, the patient complained of pain in the right side of the abdomen and chest, and shortness of breath. Her chest x-ray revealed pleural effusion on the right side. Ultrasonography showed internal genitalia and pelvic endometriosis.

Both chest computed tomography (CT) and magnetic resonance imaging (MRI) have also been used for the diagnosis of TE [2]. However, it is important to know that radiological abnormalities are transient [4]. The first line imaging method is CT, but it is poorly specific [16]. A CT scan may show endometrial implants (as hypo-attenuating areas), ground-glass infiltrates, single or multiple nodular lesions or bullous formations [2,3]. The main role of chest CT is to rule out other pulmonary diseases [16]. MRI is superior to chest CT in detecting TE [7]. It demonstrates a hyperintense lesion of endometriosis on T1- and T2-weighted images [17]. In our case, a CT scan of the chest, abdomen and pelvis was performed. It revealed a small amount of air and heterogenic fluid in the right side of the chest and abdomen, heterogenic nodules in the uterus, solid density masses in the lower part of the abdomen.

A pleural fluid cytologic examination is rarely helpful [10]. The role of bronchoscopy in the diagnosis of endometriosis is also limited, because most pathologic features are located in the periphery [2]. Histopathological confirmation of endometriosis is also difficult: according to the literature, histopathological diagnosis may be obtained in only 1/3 of the cases [11]. VATS may be used as a gold standard for direct visualisation of endometrial lesions in the thorax and tissue biopsy [18].

The treatment options for TES are medical, surgical and combined therapy. The target of medical treatment is focused on the suppression of ovarian estrogen secretion [2,10]. For this purpose, danazol and gonadotropin-releasing hormone (GnRH) agonists are used [2,3,10].

When hormonal therapy fails, surgical treatment is suggested [2,7,12]. VATS or standard thoracotomy can be performed [7]. Some authors suggest chemical pleurodesis in the presence of pleural effusion, haemothorax and pneumothorax before major surgical procedures [10]. More aggressive surgical treatment are removal of ectopic endometrial tissue, closing diaphragmatic defects, abrading of pleural surfaces, and pleurectomy [6,18]. Ectopic endometrial implants in the lungs are removed by wedge resection or limited lung segmentectomy [7]. In our case, a successful minithoracothomy, atypical lung resection and partial pleurectomy were performed. There was no recurrence of bleeding in the chest.

4. Conclusions

The diagnosis of TES is complicated and often delayed. TES should be suspected in a reproductive age woman with exacerbating symptoms during the menstruation. A great attention should be paid to the importance of taking a thorough history (especially careful gynaecological history), a comprehensive physical and radiological examination. When possible, TES treatment should be started with medicine. If medical therapy fails, surgical treatment should be performed. Our case report shows that TES may be the cause of dangerous situation when an urgent operation must be performed. Not all clinicians know that TES sometimes might complicate to pneumothorax or massive bleeding, which might cost a patient's life. We suggest performing VATS as soon as possible for reproductive age woman with unknown aetiology of pneumothorax or haemothorax to find out the lesions in pleural cavity and start appropriate treatment on time.

Conflict of interest

None of the authors have any disclosures or conflict of interests.

Funding

None.

Ethical approval

We have a consent by the patient. We have not submitted the case to the Ethics Committee approval.

Consent

We have a consent by the patient.

Author contributions

Lina Pankratjevaite – data collection, writing the paper.

Diana Samiatina-Morkuniene – surgeon performing the operation. Data collection, writing the paper.

Registration of research studies

There is not a research so it is not registered.

Guarantor

Diana Samiatina-Morkuniene.

References

[1] N. Machairiotis, A. Stylianaki, G. Dryllis, et al., Extrapelvic endometriosis: a rare entity or an under diagnosed condition? Diagn. Pathol. 8 (2013) 194.
[2] P. Azizad-Pinto, D. Clarke, Thoracic endometriosis syndrome: case report and review of the literature, Perm J. 18 (3) (2014) 61–65.
[3] T. Haruki, S. Fujioka, Y. Adachi, et al., Successful video-assisted thoracic surgery for pulmonary endometriosis: report of a case, Surg. Today 37 (2) (2007) 141–144.
[4] S.M. Hwang, C.W. Lee, B.S. Lee, et al., Clinical features of thoracic endometriosis: a single center analysis, Obstet. Gynecol. Sci. 58 (3) (2015) 223–231.
[5] C. Nezhat, J. Main, E. Buescher, et al., Robotic-assisted management of endometriosis, in: Atlas of Single-Port, Laparoscopic, and Robotic Surgery, 2014, pp. 251–261.
[6] S.S. Nair, J. Nayar, Thoracic endometriosis syndrome: a veritable pandora's box, J. Clin. Diagn. Res. 10 (April (4)) (2016) QR04–QR08, http://dx.doi.org/10.7860/JCDR/2016/17668.7700.
[7] S. Alwadhi, S. Kohli, B. Chaudhary, et al., Thoracic endometriosis – a rare cause of haemoptysis, J. Clin. Diagn. Res. 10 (April (4)) (2016) TD01–TD02, http://dx.doi.org/10.7860/JCDR/2016/16365.7530.
[8] T. Suwatanapongched, V. Boonsarngsuk, N. Amornputtisathaporn, et al., Thoracic endometriosis with catamenial haemoptysis and pneumothorax: computed tomography findings and long-term follow-up after danazol treatment, Singap. Med. J. 56 (July (7)) (2015) e120–e123, http://dx.doi.org/10.11622/smedj.2015115.
[9] J. Joseph, S.A. Sahn, Thoracic endometriosis syndrome: new observations from an analysis of 110 cases, Am. J. Med. 100 (1996) 164–170.
[10] S. Sevinç, S. Ünsal, T. Öztürk, et al., Thoracic endometriosis syndrome with bloody pleural effusion in a 28 year old woman, J. Pak. Med. Assoc. 63 (1) (2013) 114–116.
[11] D.J. Wood, K. Krishnam, M.J. Ward, Catamenial hemoptysis: a rare cause, Thorax 48 (1993) 1048–1049.
[12] A. Augoulea, I. Lambrinoudaki, G. Christodoulakos, Thoracic endometriosis syndrome, Respiration 75 (1) (2008) 113–119.
[13] R.A. Agha, A.J. Fowler, A. Saetta, et al., The SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[14] J. Nwiloh, Diaphragmatic patch: a useful adjunct in surgical treatment of recurrent catamenial hemothorax, Rev. Port. Pneumol. 17 (6) (2011) 278–282.
[15] T. Inoue, M. Chida, H. Inaba, et al., Juvenile catamenial pneumothorax: institutional report and review, J. Cardiothorac. Surg. 10 (2015 Jun 13) 83.
[16] P. Rousset, C. Rousset-Jablonski, M. Alifano, Thoracic endometriosis syndrome: CT and MRI features, Clin. Radiol. 69 (3) (2014 Mar) 323–330.
[17] G. Picozzi, D. Beccani, F. Innocenti, et al., MRI features of pleural endometriosis after catamenial haemothorax, Thorax 62 (8) (2007 Aug), 744–744.
[18] M. Peterzan, T. Reynolds, K. Dulay, et al., Thoracic endometriosis syndrome manifesting as atraumatic haemothorax causing difficult ventilation under general anaesthesia, BMJ Case Rep. 2012 (December) (2012), http://dx.doi.org/10.1136/bcr-2012-007206, pii: bcr2012007206.