Abstract. Endometriosis is an insidious, silent disease often associated with symptoms such as pelvic pain and intense cramps, irregular uterine bleeding or dyspareunia. Some patients may develop over time chest pain, rapid heartbeat, pain lying down, dizziness, shortness of breath, abdominal pain, hemoptysis which occur periodically a few days before or/and during menstruation [1]. Thoracic endometriosis is a chronic disease and leads to numerous complications [2]. Because of its rarity, it may be overlooked, resulting in a delay in diagnosis. Imaging tests such as computed tomography and magnetic resonance imaging, immunohistochemical pleural fluid analysis can confirm the presence of hormone receptor-positive endometrial glands and stroma [3].

Keywords: endometriosis, pulmonary endometriosis, catamenial hemoptysis, thoracic endometriosis, menstrual cycle
1. Introduction

Thoracic endometriosis may locate in the pleura, lung, parenchyma, airways, and/or diaphragm [4]. It is said that 33% cases of lung collapse in females were the results of thoracic endometriosis [5]. The syndrome of thoracic endometriosis is characterized by four main entities, such as catamenial pneumothorax, catamenial haemothorax, catamenial haemoptysis and lung nodule [6]. Several hypotheses had been proposed to explain the mechanisms of occurrence of thoracic endometriosis including Sampson’s retrograde menstruation theory, Meyer’s coelomic metaplasia theory, Halban’s lymphovascular microembolization theory. Unfortunately, at the moment no guidelines exist to enhance the recognition and treatment of thoracic endometriosis. Some studies suggest that immunohistochemistry estrogen/progesterone receptors and CD10 - positive might be necessary for diagnosis. In addition, pleural fluid analysis may also be helpful in confirming the diagnosis [7][8][9].

2. Purpose of this work

This research aimed to analyse studies concerning thoracic endometriosis, summarize the main features of the disease, discuss about diagnostic methods and the latest treatment to avoid late diagnosis and serious complications in women affected by endometriosis.

3. Materials and Methods

A comprehensive literature review using electronic database Pubmed, was conducted. The review was limited to sources in English language. We considered articles published from July 2010 to March 2021. Keywords such as : “endometriosis”, “pulmonary endometriosis”, “thoracic endometriosis”, “catamenial hemoptysis”, and various combinations of the above were used. Publications were selected if they related to studies concerning case reports of thoracic endometriosis, diagnostic and treatment paths. In addition, we manually reviewed the references for each article to find potentially missed studies. Besides, we identified 180 articles that were related to topics of interest. Finally, 27 studies were selected for analysis.

4. Clinical manifestations
In the literature one of the most frequently described clinical manifestations of thoracic endometriosis is catamenial pneumothorax which was first described by Maurer in 1958 [10]. It is diagnosed if the symptoms occur 24 hours before or 72 hours after the onset of menstruation. Patients often manifest symptoms such as chest pain, shortness of breath, dizziness [11]. In the vast majority of cases air in the pleural cavity accumulates on the right side, favored the distribution in the lower lobe compared to the upper or middle lobes [12]. Catamenial pneumothorax is some patients may be the first symptom of this disease and it does not have to occur monthly. The second form of the disease is catamenial hemothorax. The main symptom is prolonged worsening breathlessness. Bilateral catamenial hemothorax is rare. In 85% of patients it is located on the right side of the lungs. Hemoptysis is a medical term which describes coughing up of blood from the bronchi, larynx, trachea, or lungs. It is featured in many diseases including tuberculosis, cancer, aspergilloma, cystic fibrosis or anti-glomerular basement membrane disease [13][14]. In rare cases intermittent hemoptysis may coincide with menstrual periods in women affected by thoracic endometriosis which was first mentioned in 1954 by Alan Brews. Some studies describe that approximately 7% of women who suffer from endometriosis manifest catamenial hemoptysis [15].

Fig. 1. – Clinical manifestations of thoracic endometriosis
There has been no report of a patient with catamenial hemoptysis with massive or fatal hemoptysis. The severity of hemoptysis may be influenced by the amount of endometrial tissue and the biological activity of this tissue [16][17].

5. Theories to explain an occurrence of thoracic endometriosis

The presence of intrathoracic endometrial implants has not fully been explained. In many studies three main theories to explain occurrence of thoracic endometriosis are distinguished [18]. First is the theory of coelomic metaplasia, where pleural and peritoneal structures share a common mesothelial origin as endometrial tissue. In an invitro experimental model of endometriosis using human ovarian surface epithelium cells has shown evidence that endometriotic lesions can arise by a process of metaplasia from the ovarian surface epithelium. If ovarian surface epithelium and ovarian stromal cells are ovarian surface epithelium and ovarian stromal cells, the ovarian surface epithelium cells is formed a lumen structure, surrounded by endometrial stromal cells with an epithelial mesenchymal structure. The second theory describes transplant of endometrial tissue through vascular or lymphatic embolization. The theory of retrograde menstruation – in which uterine endometrium after menstruation may become overreactive and trapped outside the uterus by reaching the pelvis via transtubal retrograde flow. Spread to thoracic structures would occur through transdiaphragmatic migration from the pelvis to the right subdiaphragmatic area through the right paracolic gutter [19][20].

6. Available diagnostic methods

Thoracic endometriosis is still considered to be a mysterious disease and difficult to diagnose. Approximately 23% of women with extra-pelvic endometriosis do not have lesions in the pelvic [22]. What is more 68% of patients are initially misdiagnosed with another condition [23]. Diagnostics is a combination of clinical, cytological and histological findings. The radiologist can play a critical role in preventing a delayed diagnosis by suggesting this condition in the appropriate clinical situation. Chest CT may reveal hypo- or isoattenuating diaphragmatic endometrial implants. Screening of the posterosuperior diaphragm in particular is recommended, as endometrial implants have a propensity to deposit in this location.
CT findings may also include focal pleural thickening, bullae/cyst formation, ground-glass opacities or consolidation suggesting hemorrhage. MR possesses excellent contrast resolution and is helpful in detecting blood products. Pleural and diaphragmatic implants may manifest as T1-hyperintense cystic lesions, reflecting their hemorrhagic components, and will also appear hyperintense on T2-weighted sequences [24]. Identification of hormone receptor-positive endometrial glands and stroma on pathologic evaluation helps cement the diagnosis of thoracic endometriosis. Fluid/blood collected from the pleural space can be submitted for cytologic examination. Immunohistochemical staining for estrogen receptor and progesterone receptor is observed in both endometrial epithelial and stromal components. The CD10+ marker can be used to highlight endometrial stromal cells [26][27].

Fig. 2. Pleural fluid content and possible markers in thoracic endometriosis

Although thoracic endometriosis syndrome occurs infrequently, having an awareness of this entity can help prevent delay in diagnosis, allow timely institution of therapy, and possibly avoid future complications.

6. Conclusions

– Thoracic endometriosis should be suspected in a reproductive age woman with exacerbating symptoms during the menstruation
– CT and MR can help to identify lesions suspicious of endometriosis
– The key to the diagnosis are the catamenial symptoms, so a thorough clinical history is essential to promptly reach the correct diagnosis
References:


