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# Catamenial pneumothorax caused by thoracic endometriosis

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ABSTRACT

*Background:* Thoracic endometriosis syndrome is a rare form of extrapelvic endometriosis characterized by the presence of functioning endometrial tissue in pleura, lung parenchyma, and airways, associated with a high rate of infertility.

*Case Report:* We have reported a case of successful management and treatment of thoracic endometriosis syndrome that occurred in a 37-year-old female patient. She underwent thoracoscopic resection of the lesion. During follow-up, we revealed the recurrence of a previously surgically treated thoracic endometriosis. She was initially treated with a gonadotropin-releasing hormone agonist; subsequently this was replaced by a prophylactic treatment with Dienogest.

*Conclusion:* The diagnosis of thoracic endometriosis is challenging. The first line of treatment is medical, whereas the surgical treatment is performed secondly. Moreover, surgical treatment can lead to a significant rate of recurrence, often reduced by a coadjutant medical treatment.



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## Introduction

Thoracic endometriosis syndrome is a rare and crippling form of extrapelvic endometriosis. It is a clinical entity, affecting women in their reproductive years, in which deposits of functional endometrial tissue are located within the pleura, the lung parenchyma, and airways [1]. Thoracic endometriosis manifests itself through various clinical presentations such as catamenial pneumothorax (PNX) (73%), catamenial hemothorax (14%), catamenial hemoptysis (7%), and lung nodules (6%) [2]. Pelvic endometriosis is associated with 30%-50% of cases. Predisposing factors are still unknown, but a systemic immune alteration probably plays a role in the ectopic presence and persistence of endometrial tissue, although this hypothesis has yet to be clearly established [3–9]. Symptoms always appear contemporaneously with the menstrual period: the typical clinical manifestation is catamenial PNX, generally observed in about 3%-6% of spontaneous PNX, most of which involving the right side. Moreover, a history of hemoptysis during the menstrual period could be a strong indicator of pulmonary endometriosis [10]. Despite its poor specificity, high-resolution computed tomography (HRCT) of the thorax during the menstrual period remains the primary imaging method to support the diagnosis. It is capable of revealing admonishing lesions as ground-glass opacities or nodules [11]. Magnetic resonance imaging (MRI) is considered more sensitive than computed tomography (CT) in blood detection during menstruation (hyperintense in T2 gradient echo), but less in spatial resolution compared to CT [12]. Bronchoscopy effectively isolates lesions located in the airways, but it cannot be used to detect the lesions located in the lung parenchyma and pleura [13]. Pulmonary endometriosis can be treated medically or surgically. Medical therapy consists of suppression of ectopic endometrium activity by progestins, danazol, or gonadotropin-releasing hormone (GnRH) analogs [10–15]. Surgical management is performed when medical treatment fails and consists of endometrial tissue removal through video-assisted thoracoscopic surgery (VATS) or open surgery. Chemical pleurodesis could be performed in cases of catamenial PNX or hemothorax [16]. We have reported 1 case of thoracic endometriosis with recurrent episodes of catamenial PNX, associated with both pulmonary and extrapulmonary nodules and severe abdominal endometriosis.

## Case report

The patient was informed of all procedures she was to undergo, signed a consent allowing data collection for research purposes, and gave full approval for the report and publishing of the case. This case is in accordance with the Helsinki Declaration, in accordance with the Consensus-based Clinical Case Reporting Guideline Development (http://www.equator-network.org/) and the Committee on Publication Ethics (COPE) guidelines (http://publicationethics.org/), and approved by the Institutional Review Board (IRB) of the university hospital in which it was reported. A 37-year-old woman was admitted in November 2010 to our Institute with acute breathlessness, shortness of breath, chest pain, and cough typically arising during menstrual period. Initially, she had been diagnosed with serious pelvic, intestinal, bladder and abdominal wall endometriosis following a previous emergency operation that was performed because of a ruptured ovarian cyst (corpus hemorrhagicum cyst) and acute intestinal obstruction. Her menarche occurred 12 years ago, and since then she complained about severe symptoms of dysmenorrhea evaluated with visual analog scale (VAS = 9), dyspareunia (VAS = 8), dyschezia (VAS = 7), and chronic abdominal pain (VAS = 4). She reported a history of recurrent chest pain non-specifically treated and underwent multiple blood and hormone tests during this period with no results. The patient had a family history of endometriosis that occurred also in her mother, sister, aunt, cousin, and grandmother. Despite several efforts, she was unable to become pregnant. During our physical evaluation, asymmetric thorax movement and decreased breathing sound in her left hemithorax were observed. Vital parameters were stable (blood pressure 134/84 mm Hg; heart rate 95 bpm; oxygen blood saturation of 95%). A chest x-ray was performed revealing a PNX associated with areas of ground-glass haze in the anterior basal segment of the left lung, in a subpleural localization (Fig. 1). A subsequent CT scan confirmed the same lesions. A simultaneous abdominal evaluation was performed to evaluate the resumption of endometriosis disease. The patient was suffering pelvic discomfort during the gynecological evaluation, and a pelvic ultrasound (US) indicated the presence of multiple outbreaks with typical endometriosis US features of cystic ground-glass lesions involving the contralateral ovary, which had previously been operated on. Moreover, CA-125 serum was high. The patient underwent pleural and pulmonary lesion resection: mono-port VATS was performed and a macroscopically millimetric brownish and white-scattered lesion was discovered, representing small endometriosis implants into the pleural cavity. We also found emphysematous blebs and bullae matching with the anterior basal segment of the left lung where a gentle resection of lung parenchyma was performed. Histologic examination revealed the presence of endometrial tissue with glandular and stromal structures positive for estrogen receptors, which confirmed the diagnosis of pulmonary endometriosis. Consequentially, the previous catamenial episodes of PNX were linked to thoracic endometriosis. PNX was treated using chemical pleurodesis, and after surgical treatment the patient underwent medical therapy with GnRH agonists. The patient underwent a follow-up with thorax HRCT performed a month later during her menstrual cycle. Successively, an HRCT was planned 2 and 5 years later. In 2015, the patient returned to our Institute showing the same symptoms of breathlessness, cough, and pain located in the left side of the chest associated with umbilical pain. She underwent a chest MRI revealing the presence of a recurrent nodular lesion in located subpleura in the left lung, which could suggest endometriosis. A chest CT scan detected small nodules with ground-glass opacity in the left lung (Fig. 1), confirming the suspected diagnosis. Moreover, an extrapulmonary localization was detected as a ground-glass nodular lesion in the paracardiac area (Fig. 1). According to the complex thorax involvement, a fully medical treatment was performed with GnRH agonists (Enantone 3.75 mg) until the remission of symptomatology. To prevent new thorax endometriosis episodes and according to