



Thoracic endometriosis: clinicopathologic updates and issues about 18 cases from a tertiary referring center☆



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ABSTRACT

Thoracic endometriosis is defined as the ectopic presence of endometrial glands and stroma in lung or pleura and constitutes an uncommon cause of spontaneous pneumothorax in nonsmoker women in childbearing age. From 2000 to 2014, 18 (7.3%) women of 246 had a histologically proven endometriosis-related pneumothorax. The examination of thoracic samples was performed on formalin-fixed, paraffin-embedded hematoxylin and eosin-stained slides. Immunohistochemistry was carried out on all samples. The pathologic findings included endometriosis foci appearing as a triad of endometrial glands, stroma, and hemosiderin-laden macrophages in 8 patients. In the remaining 10 patients, minute nests of endometrial stroma were recognized at histologic examination and immunohistochemistry. One patient displayed diaphragmatic endometriosis and pulmonary lymphangioliomyomatosis. Thoracic endometriosis is an uncommon cause of recurrent pneumothorax necessitating histologic confirmation. The histologic diagnosis of endometriosis may be challenging on small pleural or lung biopsies because endometriosis mostly appears as minute nests of endometrial stromal cells. Here, we present our experience on thoracic endometriosis: considering frequent interpretation difficulties of biopsy samples, we propose a multidisciplinary diagnostic algorithm.

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1. Introduction

Endometriosis is a disease characterized by the ectopic dislocation of endometrial components outside the uterine cavity. Ectopic endometrium can be found in abdominal scar, breast, lung, and the pleural cavity. Pelvic endometriosis constitutes the most usual presentation of the disease, clinically characterized by abdominal pain, dysmenorrhea, and dyspareunia [1]. To the best of our knowledge, there are 3 theories about the pathogenesis of thoracic endometriosis [2]. The Ivanoff theory states that pleural tissue may undergo “endometrial metaplasia” by reason of its embryonic origin from the coelomic cavity [1]. A second theory deals with the trans diaphragmatic crossing of endometrial tissue and subsequent implantation into the pleural cavity [2,3]. The third theory involves the hematogenic migration of endometrium after surgical procedure [4]. The presence of functional endometrium in the lung or

pleura is quite uncommon and may cause hemoptysis in case of bronchial location or spontaneous pneumothorax in case of pleural or diaphragmatic involvement [5]. The typical patient profile is that of a woman in reproductive age with enduring symptoms of chest pain, cough, and dyspnea [6–9]. Although most cases revealed themselves because of the occurrence of symptoms in association with the menstrual cycle, some cases “escape” this standard, resulting in a more difficult diagnosis [10,11]. Alifano et al [11] described a recent series of thoracic endometriosis presenting with pneumothorax manifesting in the intermenstrual period which they defined as *non-catamenial endometriosis-related pneumothorax*. Pathologists are usually confronted with thoracic endometriosis by analyzing pleural biopsies or bronchial biopsies. Classic histopathologic features of endometriosis are represented by the “triad” of endometrial glands, stroma, and hemosiderin-laden macrophages [8]. The recognition of these elements is not always achieved on small tissue specimens; in addition, the presence of glandular elements in pleura or diaphragmatic biopsies may result in misleading. A well-characterized study providing clinical and comprehensive histologic assessment of endometriosis in pleural/or lung biopsy has not been undertaken until very recently. We retrospectively discuss the pathologic issues about 18 cases of thoracic endometriosis clinically presenting as spontaneous and/or recurring pneumothorax in women without known underlying lung disease. Eventually, we propose a diagnostic algorithm, for the management of thoracic biopsies in patients with suspicion of endometriosis.

Abbreviations: PNX, pneumothorax; CT, computed tomography; LAM, lymphangioliomyomatosis; BHD syndrome, Birt-Hogg-Dubé syndrome.

☆ The authors declare no conflict of interest.

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2. Material and methods

This study was approved by the Scientific and Ethical Board of the Institution. A total of 18 cases of histologically proven thoracic endometriosis were retrieved from the Pathology Department's archive from 2000 to 2014.

2.1. Immunohistochemistry technique

Hematoxylin and eosin (H&E)-dyed slides and immunohistochemistry stains were entirely re-viewed by 3 specialized thoracic pathologists (M.R.G, P.D.M., and V.T.M.). Standard immunohistochemistry were available for all cases and incorporated a panel of the following antibodies: estrogen receptor (ER; Microm clone SP1 Lab Vision Corp Fremont California USA), progesterone receptor (PR; Microm clone SP-2 Lab Vision Corp Fremont California USA), CD10 (Diagomics clone 56C6 Zytomed System GmbH Berlin Germany), HMB45 (Clinisciences clone HMB45 Lab Vision Corp Fremont California USA), and SMA (Microm Clone 1A4 Lab Vision Corp Fremont California USA). Immunohistochemistry was performed on 4- μ m-thick tissue sections from formalin-fixed, paraffin-embedded biopsy specimens. Secondary antibodies were represented by biotinylated goat polyvalent secondary antibody Ultravision system (Thermo, Lab Vision Corp Fremont California USA). The chromogenic substrates (chromogen permanent AEC kit Diagomics) were counterstained by hematoxylin and then analyzed at light microscopy.

3. Results

During the 14-year study period, a total of 1169 patients with spontaneous pneumothorax were referred to our institution. Two hundred forty-six patients were women. The mean age was 32.5 years (range, 16–54 years). A total of 185 (75.2%) had history of tobacco consumption. Eighteen (7.3%) of the 246 women were diagnosed as having thoracic endometriosis. In the 18 patients with endometriosis, a total of 41 presurgical episodes of pneumothorax were recorded. The details of the demographic and the clinical features of the 18 patients are showed in Table 1. The presenting symptoms included chest pain, dyspnea, and cough. Pain was right sided in all patients, and chest x-ray showed pneumothorax in all cases. In 9 patients, the respiratory symptoms were not clearly related with the onset of menses. Pleuroscopy revealed brown nodules or plaques on pleural surface in 4 patients and on diaphragm in 14 patients.

3.1. Histologic findings

Histologic findings are summarized in Table 2. Inflammation was constantly found in surgical pleural biopsies. The inflammatory cells were mostly represented by lymphocytes, plasmocytes, and macrophages.

Table 1
Clinical features of the patients enrolled in the study

Age (y), mean \pm SD	32.5 \pm 8.23
Notion of pelvic endometriosis	2
C-PNX	9
Non-C-PNX	9
Clinical presentation	Pneumothorax (all)
Smoke	1 (passive tobacco consumption) 12 (tobacco = 0) 5 (tobacco consumption N/A)
Chest side	
- Right	18
- Left	none
Pleural (visceral/parietal) nodules	4
Diaphragmatic anomalies (perforation/brown plaques)	14

Abbreviations: PNX, pneumothorax; C-PNX, catamenial pneumothorax.

Table 2
Histologic findings of thoracic endometriosis

Diaphragmatic endometriosis	14
Parietal pleura endometriosis	2
Visceral pleura endometriosis	2
Endometriosis features	
- Endometrial glands + stroma	8
- Stroma only	10
Hemorrhagic changes	11
Pulmonary blebs	6
Scarring of lung parenchyma	8
Inflammation changes	18

The mesothelial lining was frequently injured and exhibited reactive changes; in some biopsy samples, it was lost and replaced by fibrin. The typical histologic triad including endometrial glands, stroma, and hemosiderin-laden macrophages was recognized in 8 cases. The endometrial glands and stroma were mostly found in the subpleural connective tissue, usually associated with inflammatory cells (Fig. 1a). The phenotype of endometrial glands was that of proliferative endometrium. Secretory-type changes were not documented in our series. Stroma incorporated bland spindle cells, thin-walled capillaries, and extravasated red blood cells, usually in close association with hemosiderin-laden macrophages (Fig. 1b-d). Sometimes, the stroma appeared less cellular because of edematous changes. In our series, endometriosis involved mostly diaphragm (n = 14), whereas pleura was involved in 4 cases. Fibrosis with scarring of lung parenchyma was documented in 8 cases (Fig. 1e). In a half of cases, pleural endometriosis appeared as nests of spindle cells, without glandular elements (Fig. 1f). Immunohistochemistry (ER and PR) proved the hormone-dependent nature of these elements, which we interpreted as endometrial stromal foci. One single case was characterized by the association of diaphragmatic endometriosis and pulmonary lymphangioliomyomatosis (Fig. 1g, h). Diaphragmatic anomalies were noticed in most cases by surgeon during pleuroscopy. These consisted in brown nodules and zones of leakage (Fig. 2).

3.2. Immunohistochemistry findings

The diagnosis of pleural endometriosis on small tissue biopsy is challenging and requires immunohistochemistry. The panel of antibodies we performed on pleural biopsies included ER, PR, CD10, HMB45, and SMA. In our series, the endometrial glands and stroma were highlighted by ER and PR immunostainings. Minute nests of endometrial stroma were disclosed by hormone receptors and CD10 (Fig. 3a, b). HMB45 and SMA identified lymphangioliomyomatosis spindle cells in a lung cyst in 1 case (Fig. 3d, e).

4. Discussion

Recurrent pneumothorax is a usual manifestation of thoracic endometriosis because endometriosis implants mostly involve the diaphragm and the parietal pleura. Diverse hypothesis have been advanced to explain the pathogenesis of endometriosis related pneumothorax: spontaneous rupture of blebs [5], shedding of endometrial implants of visceral pleura [9], and the transdiaphragmatic crossing of air from the genital tract during menses [9]. The latter hypothesis is currently preferred [9]. A more unusual manifestation of thoracic endometriosis is hemoptysis, which signs the involvement of the bronchial tree [9]. In this setting, the bronchoscopic examination usually reveals pin-like mucosal lesions, but the histologic analysis of bronchial biopsies is usually nonconclusive. In our study, all patients presented with pneumothorax, without bronchial tree involvement. Even if the improvements in imaging and surgery enable a better recognition of endometriosis, the real frequency of this disease is still unclear. Over a period of 14 years, we found that 7.3% of women who, undergone surgery for pneumothorax in our institution, had thoracic endometriosis. Shearin and colleagues

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