INTERNAL MEDICINE

□ CASE REPORT □

# Pulmonary Endometriosis which Probably Occurred through Hematogenous Metastasis after Artificial Abortion

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## Abstract

Pulmonary endometriosis (PEM) is a rare disease characterized by the proliferation of ectopic endometrial tissue in the lungs, which presents as catamenial hemoptysis. A 20-year-old-woman was admitted for repeated hemoptysis. Chest CT revealed a ground-glass opacity that appeared consistently with her menstrual cycle. Our detailed inquiry revealed a history of artificial abortion, which was followed by the use of oral contraceptives and catamenial hemoptysis after the discontinuation of these medications. Surgical removal was performed and histopathological examinations confirmed PEM. This clinical course suggested hematogenous metastasis. An inquiry regarding the patient's history of uterine procedures and use of oral contraceptives was suggestive for the diagnosis of this disease.

**Key words:** pulmonary endometriosis, artificial abortion, hematogenous metastasis, oral contraceptives

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## Introduction

Pulmonary endometriosis (PEM) is a rare disease characterized by the proliferation of ectopic endometrial tissue in the lungs, which presents as catamenial hemoptysis. Its clinical profiles and etiology remain undetermined and obscure (1). However, there are two pathophysiological theories hematogenous metastasis and intraperitoneal migration that explain how endometrial tissue metastasizes to the lung (s). We herein present a case of PEM that developed after artificial abortion and the discontinuation of oral contraceptives and suggest the occurrence of hematogenous metastasis of endometrial tissue.

## Case Report

A 20-year-old woman was admitted to a nearby clinic with hemoptysis in September 2015. One month later, she was admitted to another hospital with a recurrence of hemoptysis. At that time, chest computed tomography (CT) revealed ground-glass opacity (GGO) in the right anterior basal segment (Fig. 1), and a bronchoscopic examination revealed a red, elevated lesion at the inlet portion of the anterior basal bronchus of the right lung (Fig. 2). In November 2015, she visited the hospital with a recurrence of hemoptysis. The hemoptysis only improved with the use of hemostats. Two weeks after the first recurrence of hemoptysis, the patient was admitted to our hospital for the further evaluation of her disease.

At admission, 2 weeks after her last menstruation, the patient had no symptoms. Physical examinations and laboratory investigations were unremarkable. Chest CT revealed the disappearance of the GGO in the right lower lobe. Enhanced CT revealed no abnormal blood vessels that could cause hemoptysis. A careful inquiry determined that she had undergone an artificial abortion at 18 years of age and had subsequently begun taking low-dose pills for contraception. This medication was continued until 1 month prior to the appearance of the hemoptysis (Fig. 3). PEM was suspected based on the examination results and patient's history of catamenial hemoptysis after the discontinuation of oral con-

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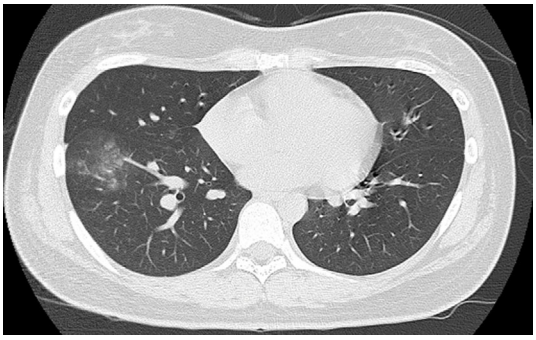

**Figure 1. Chest computed tomography at the patient's first admission. Chest computed tomography at the patient's first admission revealed ground-glass opacity in the right anterior basal segment.**

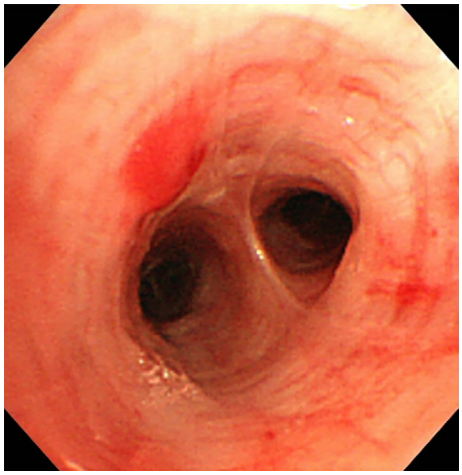

**Figure 2. Bronchoscopy at the patient's first admission. Bronchoscopy revealed a red, elevated lesion at the inlet portion of the anterior basal bronchus of the right lung.**

traceptives. Although we prescribed low-dose pills again for the prevention of hemoptysis, the patient suspended the use of these pills due to the side effects; subsequently, her catamenial hemoptysis continued. In March 2016, she underwent right anterior basal segmentectomy. During this operation, no abnormalities were detected on the surfaces of the visceral and parietal pleura and diaphragm. A histopathological examination of the resected specimen revealed the existence of endometrial glands that were immunohistochemically-positive for estrogen receptor and PAX8 and endometrial stroma that was positive for estrogen receptor and CD10 (Fig. 4). Additionally, bleeding and hemosiderosis were seen in the alveoli around the endometrial tissue. The pathological diagnosis was PEM. Her hemoptysis did not recur for 4 months after surgery without the administration of oral contraceptives.

## Discussion

The present case involves two notable clinical findings. First, the specific clinical features of PEM have rarely been reported in prior publications. PEM rarely presents with all of the specific clinical features, such as catamenial hemoptysis, changes in CT findings that appear consistently with the menstrual cycle, and artificial abortion as a risk factor. Thus, this is regarded as an exemplary case. Second, this case suggested that an artificial abortion played an important role as a risk factor for endometriosis, and indicated that metastasis of endometrial tissue via the blood vessels was involved in the development of PEM.

PEM is a rare form of extrapelvic endometriosis. Several clinical features of PEM have been reported; however, all of the features are not generally observed in each case. Thoracic endometriosis encompasses both PEM and pleural endometriosis. Most patients with thoracic endometriosis present with catamenial pneumothorax (73%) which is mainly caused by pleural endometriosis; other thoracic endometriosis patients present with catamenial hemothorax (14%). Catamenial hemoptysis, which is mainly caused by PEM, has been reported in 7% of cases of thoracic endometriosis (2). Uterine procedures such as curettage and Caesarean section are regarded as risk factors for PEM (3). The chest radiographic findings for this disease have been non-specific and vary according to the menstrual cycle; however, CT may reveal ill- or well-defined increased opacities, nodular lesions, and thin-walled cavities or bullous formations that change in appearance during the menstrual cycle (4). Histopathologically, endometrial stroma and glands were found in the lung specimen that was excised during menstruation. Surgical removal is not performed in all cases; instead, PEM is often clinically diagnosed. This case included many of the previously reported clinical features of this disease and was therefore regarded as an exemplary case. Additionally, the diagnosis of this disease typically requires time because various etiologies such as infection, malignancy, vasculitis, and drug-induced pulmonary alveolar hemorrhage are considered in the differential diagnosis of hemoptysis. However, we were able to diagnose the present case at an early stage because it featured several specific findings.

This case indicated that endometrial tissue metastasized via venous or lymphatic channels in response to an artificial abortion. Furthermore, the clinical course of this patient suggests that low-dose contraceptive pills suppressed the manifestation of PEM, which and was considered useful for elucidating the pathophysiology of PEM. The microembolization theory and the intraperitoneal migration theory are two theories that explain metastasis into the thoracic cavity. Both theories suggest that endometrial tissue is transported from the pelvis to the lung(s) through vascular/lymphatic channels or via metastatic implantation by the retrograde travel of endometrial tissue from the fallopian tubes to the peritoneum and then the thorax through defects in the diaphragm (5). In this case, bronchoscopy revealed an abnormal mucosal lesion, which was suspected to be PEM, while CT presented GGO without a nodular lesion. Additionally, no pleural or diaphragmatic lesion was identified; thus, the intraperitoneal theory, which involves the presence

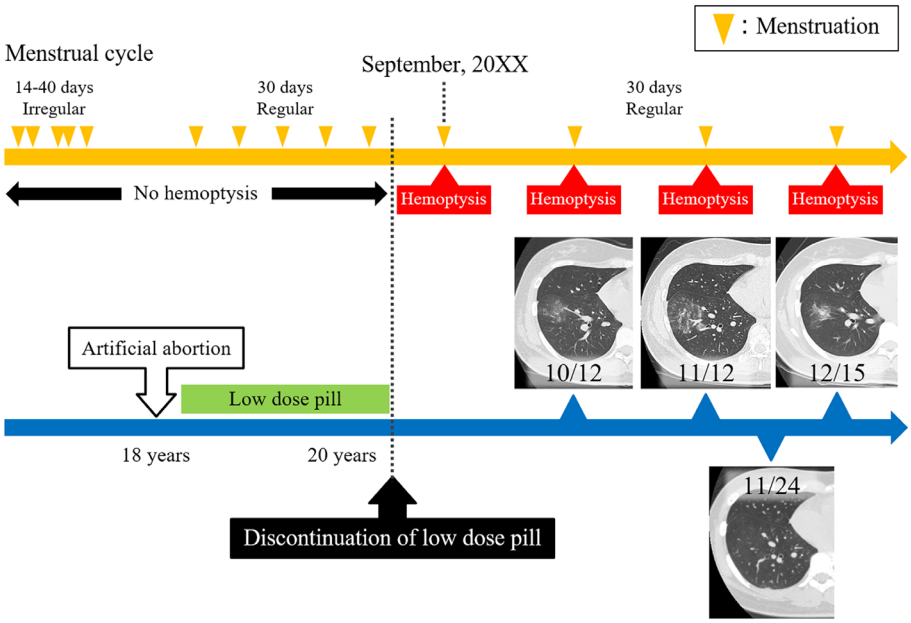


**Figure 3.** **The clinical course of the patient. The clinical course of the patient revealed catamenial hemoptysis every other month after the discontinuation of oral contraceptives.**

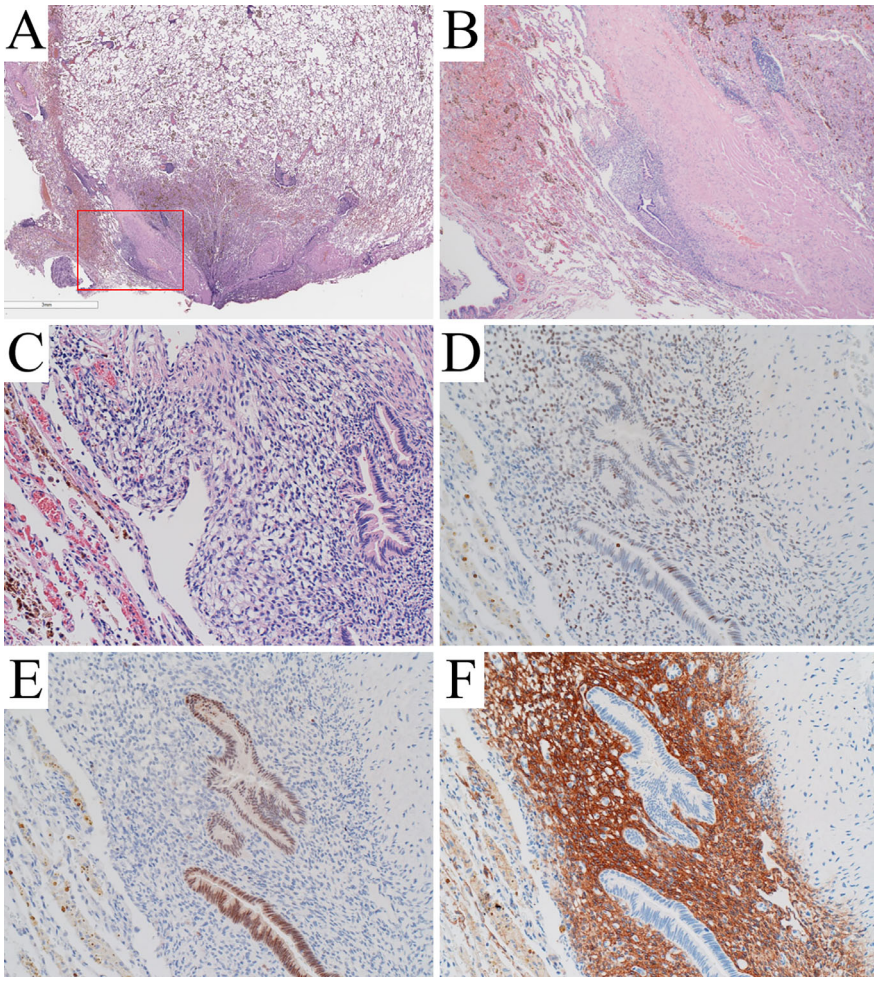


**Figure 4.** **The histopathological examination for the resected specimen. An Hematoxylin and Eosin staining resected specimen from the operation indicated the presence of endometrial tissue adjacent to a pulmonary artery in the lungs (A-C). Bleeding and hemosiderosis were observed in the alveoli around the endometrial tissue. The results of an immunohistochemical analysis revealed endometrial stroma and glands that were positive for estrogen receptor (D), endometrial glands that were positive for PAX8 (E), and endometrial stroma that was positive for CD10 (F). (A: loupe; B: ×40; C-F: ×200)**

of continuous endometrial lesions from the diaphragm to the lungs, did not apply to this case. This finding indicated a low likelihood of intraperitoneal transition. In contrast, hematogenous metastasis due to the surgical manipulation associated with artificial abortion was implicated as a cause of PEM. Additionally, this disease was suppressed by oral contraceptives. However, after the discontinuation of low-dose pills, endometriosis began to develop, resulting in catamenial hemoptysis. The micro-embolization theory was strongly supported by the patient's clinical course, which led to an early diagnosis within two months.

We herein described a case of PEM with a specific clinical course and suggested the occurrence of hematogenous metastasis. In addition to catamenial hemoptysis and CT findings that appeared consistently with the menstrual cycle, a detailed inquiry regarding the patient's history of uterine procedures and use of oral contraceptives is suggestive for the diagnosis of this disease.

**The authors state that they have no Conflict of Interest (COI).**

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