# **CASE REPORT**

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# Catamenial Hemoptysis: A Case Report and Literature Review

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# Abstract

Thoracic endometriosis is a rare form of endometriosis. It is divided into pleural and pulmonary forms, and pleural endometriosis is more common. Our patient was a 33-year-old patient who had been complaining of hemoptysis due to a menstrual cycle for 4 months and had no comorbidities. Computed tomography of the thorax during menstruation when the patient complained of hemoptysis showed a ground-glass density in the upper lobe of the right lung, which was significantly reduced on computed tomography after the end of menstruation.

**Keywords:** Catamenial, endometriosis, hemoptysis, menstrual cycle

# Introduction

Endometrial tissue growth outside the endometrium is called endometriosis and is characterized by symptoms such as infertility, dysmenorrhea, pelvic pain, and menorrhagia (1). Although endometriosis can affect up to 15% of women of reproductive age, thoracic endometriosis syndrome (TES) is a very rare condition. TES presents clinically as catamenial pneumothorax (CP), catamenial hemothorax, catamenial hemoptysis (CH), and parenchymal nodules (2). TES has two forms; pleural and pulmonary. The pleural form presents as CP, catamenial hemothorax, and chest pain, whereas the pulmonary form presents as CH and parenchymal nodules(3). The pleural form is more

common. Pulmonary endometriosis was first described by Schwarz in 1938 (1). The most common symptom of TES is CP, while the most common symptom is chest pain (3). Different theories have been proposed regarding the presence of endometrial tissues in the thorax. Some of these theories include celiomic metaplasia, i.e., the transformation of the pleural epithelium into the endometrium, while others include lymphatic and hematogenous microembolization (2,4). Here, we describe a young patient with CH who was diagnosed with computed tomography (CT) scans taken during and after menstruation. There are not many studies on this subject in the literature. For this reason, we compiled 32 articles presenting patients with CH and presented them with our case (Table 1).

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Table 1

## Studies included in the review and patient characteristics

| PUBLICATION                           | PATIENT<br>AGE | SYMPTOM    | CT FINDING                            | BIOGRAPHY            | TREATMENT                     |
|---------------------------------------|----------------|------------|---------------------------------------|----------------------|-------------------------------|
| Latters et al / 1956 (18)             | 34             | Hemoptysis | Right middle lobe ground-glass nodule | Medical abortion     | Segmentectomy                 |
| Lindenberg et al / 1975 (19)          | 43             | Hemoptysis | Right upper lobe ground-glass nodule  | Caesarean section    | Oophorectomy                  |
| Suginami et al / 1985 (20)            | 25             | Hemoptysis | Right lung ground-glass nodule        | Medical abortion     | Lobectomy                     |
| Karpel et al / 1985 (21)              | 31             | Hemoptysis | Right middle lobe nodule              | Normal               | Birth                         |
| Elliot et al / 1985 (22)              | 30             | Hemoptysis | Right lung nodule                     | Medical abortion     | Danazol                       |
| Hertzanu et al / 1987 (23)            | 32             | Hemoptysis | Right lower lobe nodule               | Medical abortion     | Progesterone                  |
| Lawrence et al / 1988 (24)            | 34             | Hemoptysis | Left lower lobe nodule                | Medical abortion     | Birth                         |
| Bateman et al / 1990 (25)             | 33             | Hemoptysis | Normal                                | Medical abortion     | Danazol                       |
| Guidry et al / 1990 (26)              | 30             | Hemoptysis | Bilateral density increase            | Medical abortion     | Progesterone                  |
| Kristiansen et al / 1993 (27)         | 24             | Hemoptysis | Right middle lobe ground-glass nodule | Normal               | Lobectomy                     |
| Joseph et al / 1994 (28)              | 30             | Hemoptysis | Pleural effusion                      | Laparatomy           | Danazol                       |
| Kuo et al / 1996 (29)                 | 31             | Hemoptysis | Right middle lobe ground-glass nodule | Normal               | Danazol                       |
| Huang et al / 2013 (30)               | 29             | Hemoptysis | Left upper lobe ground-glass nodule   | Medical abortion     | Lobectomy                     |
| Yisa et al / 2004 (31)                | 36             | Hemoptysis | Right upper lobe nodule               | Laparascopy          | İmplanon                      |
| Cassina et al / 1997 (32)             | 26             | Hemoptysis | Right upper lobe nodule               | Normal               | Wedge resection               |
| Chatra et al / 2012 (3)               | 34             | Hemoptysis | Right upper lobe ground-glass nodule  | Over kisti           | Oral contraceptive            |
| Chen et al / 2020 (33)                | 18             | Hemoptysis | Right upper lobe ground-glass nodule  | Normal               | Segmentectomy                 |
| Burdon et al / 2001 (34)              | 37             | Hemoptysis | Normal                                | Medical abortion     | Lobectomy                     |
| Nakashima et al / 2011 (35)           | 22             | Hemoptysis | Right middle lobe ground-glass nodule | Normal               | Lobectomy                     |
| Tong et al / 2019 (1)                 | 29             | Hemoptysis | Right lower lobe ground-glass nodule  | Laparoscopy          | Wedge resection               |
| Wood et al / 1993 (36)                | 32             | Hemoptysis | Right upper lobe ground-glass nodule  | Mol hidatiform       | Segmentectomy                 |
| Matsubara et al / 1997 (37)           | 19             | Hemoptysis | Left lower lobe ground-glass nodule   | Medical abortion     | GNRH agonist                  |
| Fujimoto et al / 2017 (38)            | 20             | Hemoptysis | Right lower lobe ground-glass nodule  | Medical abortion     | Segmentectomy                 |
| Gill et al / 2003 (39)                | 28             | Hemoptysis | Left upper lobe ground-glass nodule   | Laparascopy          | GNRH agonist                  |
| Lu et al / 2006 (13)                  | 17             | Hemoptysis | Left lower lobe ground-glass nodule   | Medical abortion     | Wedge resection               |
| Suwatanapongched et al / 2015<br>(40) | 38             | Hemoptysis | Right lower lobe ground-glass nodule  | Caesarean section    | Danazol                       |
| Furuya et al / 2017 (8)               | 21             | Hemoptysis | Right middle lobe ground-glass nodule | Medical abortion     | Wedge resection               |
| Aboujaoude et al / 2021 (41)          | 34             | Hemoptysis | Right upper lobe nodule               | Laparatomy           | GNRH agonist                  |
| Park et al / 2006 (42)                | 31             | Hemoptysis | Right upper lobe nodule               | Caesarean<br>section | Wedge resection               |
| Shin et al / 2014 (6)                 | 34             | Hemoptysis | Left lower lobe ground-glass nodule   | Normal               | Bronchial artery embolization |
| Yao et al / 2023 (5)                  | 19             | Hemoptysis | Right middle lobe ground-glass nodule | Normal               | Lobectomy                     |
| Kim et al / 2020 (7)                  | 26             | Hemoptysis | Right lower lobe ground-glass nodule  | Normal               | GNRH agonist                  |

## **Case Report**

A 33-year-old woman presented with hemoptysis for 4 months. Her complaints coincided with the menstrual bleeding period. Before the first episode of hemoptysis, she complained of hemoptysis with flu-like symptoms and consulted the pulmonology department. Pneumonia treatment was initiated after a ground-glass nodule was observed in the upper lobe of the right lung on thorax CT (Figure 1A). Despite pneumonia treatment, the patient presented with recurrent hemoptysis during the next menstrual bleeding. Her physical examination was normal, and she had a 13-year pack-year smoking history. She underwent ovarian cyst excision approximately 6 months ago, and no pathology was detected in current blood tests, urine tests, and respiratory tract cultures. The patient's family history included only



Figure 1A, 1B: Thoracic CT images taken during different menstrual periods

cardiologic diseases, and there was no psychiatric disorder. Thorax CT repeated during the menstrual period showed the same findings (Figure 1B). Thorax CT performed on the 14th day outside the menstrual period showed that the nodule in the upper lobe of the right lung had disappeared, and the patient was diagnosed as TES clinically and radiologically (Figure 2). Medical, interventional, and surgical treatment options for the treatment of CH were shared with the patient. However, the patient preferred bronchial artery embolization (BAE), so she was referred to the relevant unit.



Figure 2: Thoracic CT image taken 14 days after menstruation

# Discussion

Patients with TES often present with clinical findings such as recurrent chest pain, dyspnea, hemoptysis, pneumothorax, and hemothorax, and most of them are not initially diagnosed with TES. In patients presenting with recurrent hemoptysis, diseases such as tuberculosis, pneumonia, and bronchiectasis are usually initially investigated. Thorax CT findings include pneumothorax and high-density nodules (5). The theory of vascular or lymphatic microembolization may explain TES. Trauma to the uterine tissue predisposes to microembolization. Most of these patients have an obstetric and gynecologic history, which can cause uterine trauma. In a Korean study including 19 patients with catamenial hemoptysis, it was reported that 16 patients had such a history(6). Our patient also had a gynecological history. Our patient had CH, which is the pulmonary form of TES. The diagnosis of CH is usually made clinically and by excluding other causes, whereas the diagnosis of surgical patients can be made by histopathological demonstration of endometrial tissues. The clinical finding is periodic hemoptysis simultaneously with menstruation. In the report by Kim et al. 8 of 19 patients were diagnosed histopathologically and 11 were diagnosed clinically (7). Nodules can be seen on X-ray. These nodules are endometrial nodules formed as a result of the implantation of endometrial tissue into the lung parenchyma. Thoracic CT is useful for detecting ground-glass nodules and excluding other causes. In CT follow-up, the size of the nodules may change according to the menstrual cycle (6). In this patient, it was observed that the size of the nodule decreased on CT after the menstrual bleeding ended. While thoracic endometriosis is most commonly observed on the right side, there is no opinion proving this (8). In a study of 110 patients by Channabasavaiah et al., 85% of the lesions were on the right side, whereas in our patient, they were also on the right side (9). Since most pathologic lesions are located in the parenchyma in the distal bronchi, and intrabronchial lesions are usually absent, the role of bronchoscopy is limited in diagnosis (6,10). In the analysis study by Kim et al., no

intrabronchial lesion was found in 84% of patients who underwent bronchoscopy during hemoptysis attacks (7). However, Wang et al. reported that it was useful to perform bronchoscopy within the first two days after the onset of the menstrual cycle (11).

There are no guidelines for CH treatment. Patients presenting with hemoptysis usually start drugs such as transamin. Hormonal therapy is considered the first treatment option for CH. Oral contraceptives, progesterone derivatives, danazol, and gonadotropinreleasing hormone (GnRH) agonists are used in medical treatment (2). Surgical treatment is also performed due to failure of medical treatment and possible side effects. Operative techniques include wedge resection, lobectomy, pneumonectomy, and segmentectomy, and wedge resection gives better results compared to others (12). Preoperative marking can be performed with the help of CT and bronchoscopy, so the part to be resected lung can be determined more accurately. For example, Furuya et al. performed CT-guided marking with fluorescein sodium and resection of the 4th segment of the right lung (8). Lu et al. performed preoperative CTguided wire marking(13). According to the literature, spontaneous regression is rarely observed in patients who do not receive any treatment (14).

Shin et al. preferred BAE as a different method in the treatment of CH. BAE is generally used as an alternative to surgery for hemoptysis due to causes such as lung cancer, bronchiectasis, and tuberculosis (6). The procedure was first performed in 1973, and many publications have emphasized its efficacy. However, this procedure may have rare complications, such as postembolism syndrome, stenosis, tracheoesophageal fistula, spinal cord injury, transient chest pain, and an esophageal ulcer (6,15). Kervancioglu et al. reported no recurrence for 3 months after BAE in a patient with CH(16). Katoh et al. did not find any pathology in the bronchial angiography of patients with pulmonary endometriosis (17). Shin et al. found a small nodular spotting on bronchial angiography and followed the patient for 5 months without recurrence (6).

There are no guidelines for the diagnosis and treatment of thoracic endometriosis have been published. It requires early diagnosis and treatment due to pneumothorax, hemothorax, and hemoptysis. The most effective method for diagnosing the disease is to determine whether the hemoptysis coincides with the menstrual cycle as a result of good anamnesis and then to perform consecutive thoracic CT scans in periods with and without menstrual bleeding. Thus, early diagnosis and treatment will improve the patient's standard of living and enable treatment without increasing costs.

## **Conflict of Interest Statement**

There is no conflict of interest, personal or financial.

## **Consent to Participate and Publish**

Written informed consent to participate and publish was obtained from all individual participants included in the study.

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There is no financing.

### Availability of Data and Materials

Data available on request from the authors.

## **Authors Contributions**

HEY: Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Validation; Visualization; Writing-original draft.

HEC: Investigation; Validation; Formal analysis; Writing-original draft.

SEA: Investigation; Validation; Formal analysis; Writing-original draft.

RY: Data curation; Formal analysis; Invetigation; Validation; Writing- review & editing.

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