

## Case Report

# A Woman with Recurrent Hemoptysis, a Rare Etiology

Pasakorn Jitruckthai MD\*

\* Department of Medicine, Chonburi Hospital, Chonburi, Thailand

*Bronchopulmonary endometriosis is a rare clinical entity of thoracic endometriosis syndrome (TES). The diagnosis is often delayed because of high index of clinical suspicion is needed. We submit a case of 32-year-old healthy woman presenting with recurrent non-massive hemoptysis with the onset of menses for six months. Computed tomography scans of the chest revealed ill-defined ground glass opacity in superior segment of right lower lobe. Fiberoptic bronchoscopy was performed during the menstruation showed diffuse erythema along distal trachea through lobar bronchus. These findings disappeared when repeated at the end of menstrual cycle. Cytologic findings of bronchial wash suggested the evidence of old hemorrhage and the endometrial cells. After treatment with depot medroxyprogesterone acetate, hemoptysis did not occur for 12 months of follow-up. Based on clinical features and response to treatment, bronchopulmonary endometriosis was diagnosed.*

**Keywords:** Bronchopulmonary endometriosis, Catamenial hemoptysis, Thoracic endometriosis syndrome

*J Med Assoc Thai 2015; 98 (6): 616-20*

**Full text. e-Journal:** <http://www.jmatonline.com>

Cyclic hemoptysis occurring synchronously during menstruations (catamenial hemoptysis) is a rare presentation of thoracic endometriosis syndrome (TES). Fewer than 60 cases of catamenial hemoptysis are described in literature published in English<sup>(1,2)</sup>. Diagnosis of TES is often made on the basis of high index of clinical suspicion in woman with symptoms that recur with menses, so the diagnosis is often delayed because of incapability to correlate the patient's symptoms with menses.

We report a case of bronchopulmonary endometriosis presenting with catamenial hemoptysis.

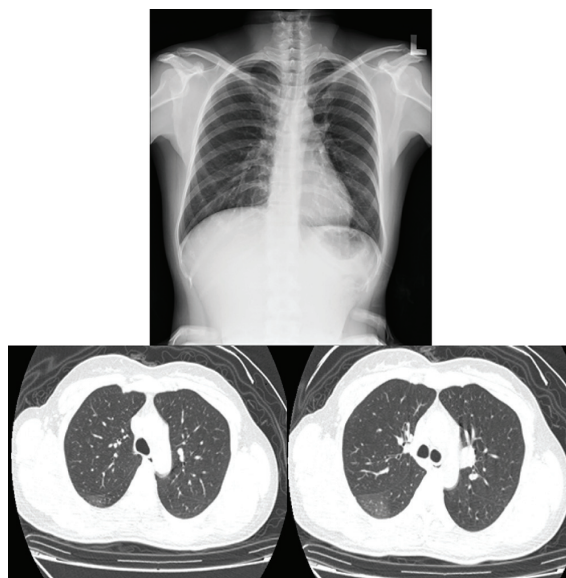
### Case Report

A 32-year-old woman (gravida 1, para 1) presented with recurrent non-massive hemoptysis with the onset of menses for six months. She described episodes of hemoptysis approximately one to two teaspoon that lasted for one week per episode during menstruation and resolved spontaneously. She had no symptoms other than hemoptysis. She was a non-smoker and denied any history of lung disease and previous pelvic manipulations or trauma.

The result of the physical examination was normal. Chest radiograph showed no abnormal findings, but computed tomography scans of the

chest revealed ill-defined ground glass opacity in superior segment of right lower lobe (Fig. 1).

Fiberoptic bronchoscopy was performed during menstruation. It showed diffuse erythema along distal trachea through lobar bronchus (Fig. 2). Bronchial wash was obtained and cytologic examinations demonstrated numerous mixed inflammatory cells, many hemosiderophages, and rows of columnar



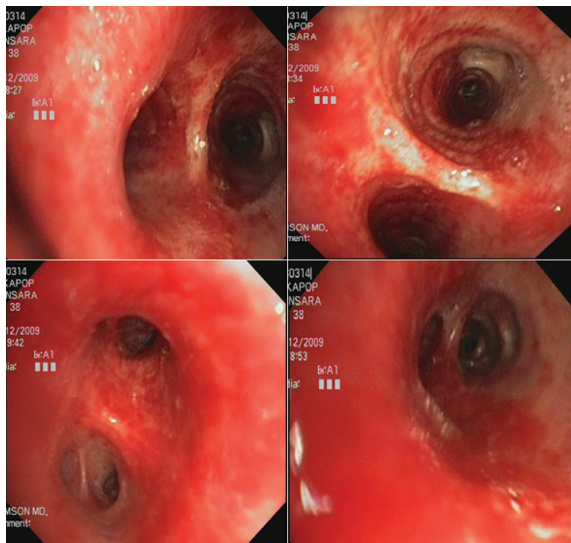
**Fig. 1** Chest radiograph show no abnormal findings and computed tomography scans of the chest demonstrate ill-defined ground glass opacity in superior segment of right lower lobe.

### Correspondence to:

Jitruckthai P, Department of Medicine, Chonburi Hospital, 69 Sukhumvit Road, Muang, Chonburi 20000, Thailand.

Phone: +66-38-931000

E-mail: Ppasakorn26338@hotmail.com



**Fig. 2** Bronchoscopic findings during menstruation show diffuse erythema along distal trachea through lobar bronchus.

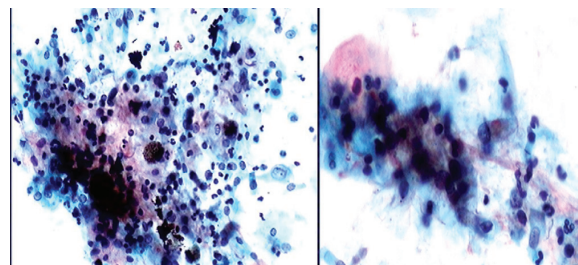
epithelium with intracytoplasmic secretion (Fig. 3). These cytological findings suggested the evidence of old hemorrhage and the endometrial cells. Unfortunately, there is not enough specimen to make immunohistochemical study for CD10 to confirm the endometrial components. Bronchial wash for acid-fast bacilli and gram stain were negative. Biopsy was not performed. Repeated fiberoptic bronchoscopy at the end of menstrual cycle showed disappearance of the former findings as described during the menstruation (Fig. 4).

Based on clinical features of recurrence hemoptysis associated with menses and dynamic change of bronchoscopic appearance with cytological result, bronchopulmonary endometriosis was diagnosed. Treatment was started with depot medroxyprogesterone acetate (DMPA) 150 mg intramuscularly. In the next month after treatment, hemoptysis did not occur. DMPA was prescribed every three months. After treatment for one year, we decided to stop medication; however, hemoptysis recurred five months after discontinuing DMPA. DMPA was restarted every three months and hemoptysis did not recur during 12 months of follow-up.

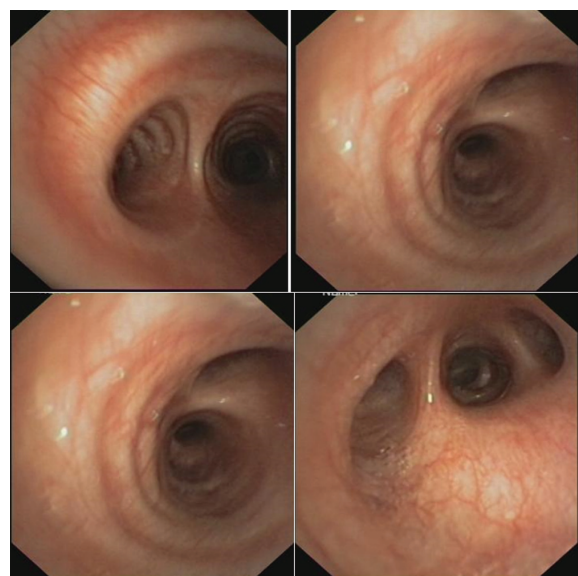
### Discussion

Thoracic endometriosis syndrome (TES) is a rare disorder characterized by the presence of functioning endometrial tissue in pleura, lung parenchyma, airways, and diaphragm<sup>(3)</sup>.

TES is a spectrum of disease, categorized into four clinical entities, catamenial pneumothorax (72% of TES), catamenial hemothorax (12-14%), catamenial hemoptysis (5-14%), and pulmonary nodule (2-6%)<sup>(1,4,5)</sup>. Hemoptysis represents the early manifestation, where the abnormality is confined to the lung parenchyma. Pneumothorax represents seeding of visceral pleura with disruption of the pleural membrane during menstruation. Finally, hemothorax, a late manifestation of TES, signaled the presence of a high burden of proliferating parietal and visceral pleura implants<sup>(1)</sup>.



**Fig. 3** Cytological examination of bronchial wash performed during menstruation demonstrated numerous mixed inflammatory cells, many hemosiderophages and rows of columnar epithelium with intracytoplasmic secretion. Some ciliated bronchial epithelial cells with reactive changes are also demonstrated.



**Fig. 4** Repeated fiberoptic bronchoscopy at the end of menstrual cycle show disappearance of the former findings as described during the menstruation.

Catamenial hemoptysis was first described as a case of hemoptysis associated with menses in pulmonary endometriosis by Lattes et al since 1956<sup>(6)</sup>. The diagnosis based on high clinical suspicion in women of reproductive age with cyclical hemoptysis during menstruation (catamenial) and exclusion of other causes of recurrent hemoptysis<sup>(7)</sup>. Diagnosis is often delayed and average duration of symptoms prior to diagnosis was 15±2.4 months<sup>(1)</sup>.

From the previous case reports of bronchopulmonary endometriosis, most patients had the history of obstetric or gynecological procedures such as dilatation and curettage, induced abortion. All cases had the suggestive clinical clue of catamenial hemoptysis. History of intermittent sharp pleuritic chest pain occurred in some cases. Chest radiographs were normal in most cases and chest CT might demonstrate nodular lesions, ground glass opacities or cystic lesion with alveolar infiltration. Bronchoscopic findings revealed multiple purplish red submucosal patches along tracheobronchial tree in some cases. Dynamic radiographic and bronchoscopic appearance during menstrual cycles strongly suggests a diagnosis of bronchopulmonary endometriosis. Histologic confirmation has been obtained in less than one-third of the previously reported case<sup>(2,8-13)</sup>.

In Thailand, only one reported case was found from the literature. This case was diagnosed by clinical suggestion of catamenial hemoptysis and exclusion other potential causes of hemoptysis. The dynamic change of bronchoscopic findings and cytologic confirmation were not demonstrated<sup>(14)</sup>.

In the present case, the diagnosis was made on the clinical history, CT findings (to localize pulmonary endometriosis and exclude other causes of hemoptysis), cyclic changes of the bronchoscopic findings, cytologic features, and together with response to the hormonal treatment. These were sufficient to diagnose bronchopulmonary endometriosis.

Pathogenesis of TES is not well understood. There are several hypotheses have been proposed. After movement of shed endometrial tissue through the fallopian tubes and into the pelvic cavity, the potential mechanism for migration of endometrial tissue from the pelvis to the thoracic cavity remains uncertain. The first hypothesis is trans-diaphragmatic movement through diaphragmatic defects. This is supported by the almost right-sided predominance of endometriotic pleural implants. The other route is hematogenous dissemination of the endometrium caused by uterine procedures. In the present case, patient denied previous

history of pelvic manipulations<sup>(3,15)</sup>. Furthermore there are another theory explain ectopic endometrial tissue develops from pluripotential cells capable of transformation into endometrial tissue with the appropriate hormonal stimulation<sup>(1,16)</sup>.

About treatment, controversy exists as to the proper treatment for catamenial hemoptysis because it has been treated with both hormonal therapy and pulmonary resection. Medical treatment including danazol, oral contraception, progestin, and gonadotropin-releasing hormone (GnRH) analogs are generally recommended as first line therapy. The aim of the medical treatment consists of suppression of ectopic thoracic endometrial tissue. Surgery is an option if the side effects of hormonal therapy are intolerable or if recurrence occurs. Endoscopic ablation with Nd-Yag laser is a new treatment option for catamenial hemoptysis in patients with centrally located tracheobronchial endometriosis<sup>(2,12,13,17,18)</sup>. In our patient, we decided to treat with Depot medroxyprogesterone acetate intramuscular every three months because of less side effect and more convenience for the patient. Hemoptysis did not recur for 12 months of follow-up.

Catamenial hemoptysis is a rare presentation of TES. A high index of suspicion should exist for women of reproductive age who present with recurrence hemoptysis synchronously during menstruation. Proper timing of bronchoscopic examination, cytologic/histologic evaluation with therapeutic trial with hormonal treatment is sufficient to warrant the diagnosis.

#### **What is already known on this topic?**

Catamenial hemoptysis is a rare presentation of thoracic endometriosis syndrome. Fewer than 60 cases are reported in the literature. The diagnosis depends on high index of clinical suspicion and cytopathologic confirmation of the diagnosis is infrequent. Furthermore, controversy exists as to the proper treatment for catamenial hemoptysis.

#### **What this study adds?**

The present case report demonstrated the typical clinical presentation of bronchopulmonary endometriosis and completed studies including chest radiograph, computed tomography scans of chest, and bronchoscopic findings during and at the end of menstrual cycle were illustrated. Moreover, cytologic examination of bronchial wash suggested the evidence of endometrial cells too.

Furthermore, treatment with intramuscular DMPA was successful. Adverse effects were not reported. This treatment may be another option for bronchopulmonary endometriosis that is effective, cheap, more convenient, and cause fewer adverse effects.

#### Acknowledgement

Dr. Kaimook Gosinthrajit, pathologist who gave the pictures of cytology from bronchial wash and described cytologic findings.

#### Potential conflicts of interest

None.

#### References

1. Channabasavaiah AD, Joseph JV. Thoracic endometriosis: revisiting the association between clinical presentation and thoracic pathology based on thoracoscopic findings in 110 patients. *Medicine (Baltimore)* 2010; 89: 183-8.
2. Inoue T, Kurokawa Y, Kaiwa Y, Abo M, Takayama T, Ansai M, et al. Video-assisted thoracoscopic surgery for catamenial hemoptysis. *Chest* 2001; 120: 655-8.
3. Nezhat C, Hajhosseini B, Buescher E, Hussein A, Hilaris GE, Sellin M. Thoracic endometriosis syndrome. In: Wetter PA, Kavic MS, Nezhat C, Winfield H, editors. *Prevention & management of laparoendoscopic surgical complications*. 3<sup>rd</sup> ed. Miami, FL: Society of Laparoendoscopic Surgeons; 2012: 1-8.
4. Joseph J, Sahn SA. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. *Am J Med* 1996; 100: 164-70.
5. Jubanyik KJ, Comite F. Extrapelvic endometriosis. *Obstet Gynecol Clin North Am* 1997; 24: 411-40.
6. Lattes R, Shepard F, Tovell H, Wylie R. A clinical and pathologic study of endometriosis of the lung. *Surg Gynecol Obstet* 1956; 103: 552-8.
7. Guidry GG, George RB. Diagnostic studies in catamenial hemoptysis. *Chest* 1990; 98: 260-1.
8. Wood DJ, Krishnan K, Stocks P, Morgan E, Ward MJ. Catamenial haemoptysis: a rare cause. *Thorax* 1993; 48: 1048-9.
9. Kim CJ, Nam HS, Lee CY, Yum HK, Yang SH, Seo KH, et al. Catamenial hemoptysis: a nationwide analysis in Korea. *Respiration* 2010; 79: 296-301.
10. Yu Z, Fleischman JK, Rahman HM, Mesia AF, Rosner F. Catamenial hemoptysis and pulmonary endometriosis: a case report. *Mt Sinai J Med* 2002; 69: 261-3.
11. Hachiya T, Okada M, Takamizawa A, Hasegawa M, Honda T, Kubo K. Catamenial hemoptysis. *Intern Med* 2003; 42: 765-6.
12. Ryu JS, Song ES, Lee KH, Cho JH, Kwak SM, Lee HL. Natural history and therapeutic implications of patients with catamenial hemoptysis. *Respir Med* 2007; 101: 1032-6.
13. Elbek O, Borekci S, Dikensoy E, Kibar Y, Bayram H, Bakir K, et al. Catamenial hemoptysis. *Tuberk Toraks* 2008; 56: 87-91.
14. Maneechotesuwan K, Chierakul N, Dejsomritrutai W, Suthinond P. A 29-year-old woman with recurrent hemoptysis. In: Maneechotesuwan K, Chierakul N, Dejsomritrutai W, Suthinond P, editors. *Pulmonary tips*. Bangkok: LT Press; 2000: 1-4. (in Thai)
15. Vinatier D, Orazi G, Cosson M, Dufour P. Theories of endometriosis. *Eur J Obstet Gynecol Reprod Biol* 2001; 96: 21-34.
16. Joseph J. Thoracic endometriosis. UpToDate [Internet]. 2013 [cited 2013 Mar 21]. Available from: <http://www.uptodate.com/contents/search>
17. Koizumi T, Inagaki H, Takabayashi Y, Kubo K. Successful use of gonadotropin-releasing hormone agonist in a patient with pulmonary endometriosis. *Respiration* 1999; 66: 544-6.
18. Terada Y, Chen F, Shoji T, Itoh H, Wada H, Hitomi S. A case of endobronchial endometriosis treated by subsegmentectomy. *Chest* 1999; 115: 1475-8.

---

## ผู้ป่วยหญิงมาด้วยอาการไอเป็นเลือดซ้ำหลายครั้ง สาเหตุที่ไม่พบบ่อย

ภาสกร จิตรรักไทย

ภาวะเยื่อโพรงมดลูกต่างที่ในหลอดลมเป็นภาวะที่พบได้ยากอันหนึ่งในกลุ่มอาการเยื่อโพรงมดลูกต่างที่ในช่องอก การวินิจฉัยมักล่าช้าเนื่องจากต้องอาศัยลักษณะอาการทางคลินิกที่ชวนสงสัย ผู้นี้พบได้ในนำเสนอผู้ป่วยหญิงอายุ 32 ปี ไม่มีโรคประจำตัวใดๆ ที่มีอาการไอเป็นเลือดซ้ำสัมพันธ์กับรอบประจำเดือนเป็นเวลา 6 เดือน ผลการตรวจคอมพิวเตอร์ปอดพบรอยฝ้าเล็กน้อยที่ปอดกลีบขวาล่าง ส่งกล้องตรวจหลอดลมระหว่างรอบประจำเดือนพบรอยแดงกระจายทั่วหลอดลมปอด และรอยแดงดังกล่าวได้หายไปในการส่องตรวจซ้ำหลังหมดรอบเดือน การตรวจเซลล์วิทยาของน้ำล้างหลอดลมพบลักษณะของเซลล์อักเสบและเซลล์เยื่อโพรงมดลูก หลังการรักษาด้วยยาฉีดฮอร์โมนคุมกำเนิด อาการไอเป็นเลือดหายไปจนถึง 12 เดือน ที่มีการติดตามอาศัยจากลักษณะทางคลินิกและการตอบสนองต่อการรักษาทำให้ผู้ป่วยได้รับการวินิจฉัยภาวะเยื่อโพรงมดลูกต่างที่ในหลอดลม

---