Catamenial hemoptysis

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ÖZET

Katameniyal hemoptizi

Nadir gözlenen bir patoloji olan katameniyal hemoptizi intrapulmoner ya da endobronşiyal alanlarda endometriyum dokusunun varlığıyla karakterizedir. Bu makalede endobronşiyal tutulum ile seyreden katameniyal hemoptizili bir olgu sunulmuştur. Yirmi iki yaşındaki hastanın öyküsünde son iki yıldır menstrüel siklus döneminde tekrarlayan hemoptizi epizodları mevcuttu. Menstrüasyonun ilk gününde yapılan bronkoskopik incelemesinde trakea 1/3 distalinde ve her iki bronş sisteminde, dokunmakla kolayca kanayan, pembe-kırmızı/vişne çürüğü renkte yama tarzında alanlar izlendi. Bu bölgelerden yapılan bronşiyal fırçalama örneklemesinin sitolojik incelemesinde endometriyum orjinli küçük küboidal hücrelerin oluşturduğu hücre grupları saptandı. Menstrüasyonun bitiminden sonra tekrarlanan bronkoskopik incelemede ise ilk bronkoskopide tespit edilen tüm lezyonların kaybolduğu görüldü. Hastaya gonadotropin-salgılatan hormon analoğu ile östrojen ve progesteron içeren hormon replasman tedavisi başlandı. Tedavi ile tam yanıt alındı.

Anahtar Kelimeler: *Katameniyal hemoptizi, endobronşiyal endometriyozis, bronşiyal fırça sitolojisi.*

SUMMARY

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Catamenial hemoptysis is a rare condition that is associated with the presence of intrapulmonary or endobronchial endometrial tissue. We describe a case of endobronchial endometriosis with catamenial hemoptysis. The patient was a 22 years-old girl presented with recurrent hemoptysis episodes for the last two years. Bronchoscopic examination was performed within first days of menses, and indicated multiple purplish-red submucosal patches in distal one third of trachea and bilateral bronchial trees that bled easily when touched. The cytological evaluation of the bronchial brushing specimens demonstrated clusters of small cuboidal cells consistent with an endometrial origin. Follow-up bronchoscopic examination at the end of the menstrual cycle revealed that the previous tracheobronchial lesions disappeared. The patient was treated with Gonadotropin-Releasing Hormone (GnRH) analogues and hormones including estrogen and progesterone therapy. Recurrent hemoptysis stopped following the medical treatment.

Key Words: *Catamenial hemoptysis, endobronchial endometriosis, bronchial brush cytology.*

Catamenial hemoptysis is a rare disease that occurs synchronously during the menstrual cyle of female patients. Intrapulmonary endometrial tissue that is present in the parenchyma and/or central airways causes cyclic pulmonary hemorrhage. Catamenial hemoptysis is still rare condition since it was first published by Lattes et al (1).

To our knowledge, there are less than 40 patients reported in the English literature, and the histopathological confirmation was obtained in only one-third of the reported cases (2-9).

Endometriosis involving trachea and/or large bronchi is a very rare condition. There are only 10 proven cases, who have been reported previously (2). The main criterion for the diagnosis is the finding of periodic hemoptysis that is synchronous with menstruation, and most of the reported cases were diagnosed on the basis of the patient's clinical history without histological confirmation (3,10-12).

In this report, we present a case of bronchial endometriosis diagnosed on the basis of clinical history and bronchoscopic evaluation including bronchial brush cytology. The disease was successfully controlled with Gonadotropin-Releasing Hormone (GnRH) analogues in addition to estrogen and progesterone therapy. Hemoptysis has not been recurred during the last three months of the treatment.

CASE REPORT

A 22 years-old, non-smoking girl presented with a two years history of recurrent hemoptysis occurring during menstrual cycles. She claimed that hemoptysis started on the first or second days of her each menstrual cycle. The volume of expectorated blood ranged from 5 to 100 mL and spontaneous resolution was always occurred by the fourth day of the cycle.

Her vital signs on admission were as follows; respiratory rate, 18 breaths/minute; arterial blood pressure, 120/80 mmHg; heart rate, 76 beat/minute; body temperature, 36.7°C; and pulse oxygen saturation, 97%. Her general physical examination was normal. Blood analysis showed that hemoglobin was 12 g/dL, white blood cell count was 9850/mm^3, erythrocyte sedimentation rate was 10 mm/hour, C-ANCA and P-ANCA were negative, serum C-reactive protein level was normal. Her chest X-ray was normal. Sputum smears were negative for acid-fast bacilli and malignant cells. She had a history of oligomenorrhea. Gynecological examination revealed no evidence of pelvic or abdominal endometriosis. High resolution computed tomography of the chest revealed rare micronodular infiltration in both lungs parenchyma along with ground glass opacities in the right lower lobe, and tubular bronchiectatic areas in a small region of the right upper lobe.

Bronchoscopic examination performed on the first day of her menstrual cycle disclosed multiple purplish-red submucosal patches in the one-third of distal trachea and bilateral bronchial trees that bled easily when touched (Figure 1). Cytological evaluation of the brushing material demonstrated clusters of small cuboidal cells consistent with an endometrial origin (Figure 2). Cultures of bronchial brushings were negative for *Mycobacterium tuberculosis* and fungi. Follow-up bronchoscopic examination at the end of her menstrual cyle on the fifth day, showed no indication of previous tracheobronchial lesions (Figure 3). A medical therapy regimen including GnRH analogues for the catamenial hemoptysis, and a low dose oral contraceptive for the protection from side effects of the GnRH analogue was started. She did not experience an episode of hemoptysis then after.

DISCUSSION

In this article, we report a case of endobronchial endometriosis, who was diagnosed by means of cytological examination of brush specimens obtained during the fiberoptic bronchoscopy.

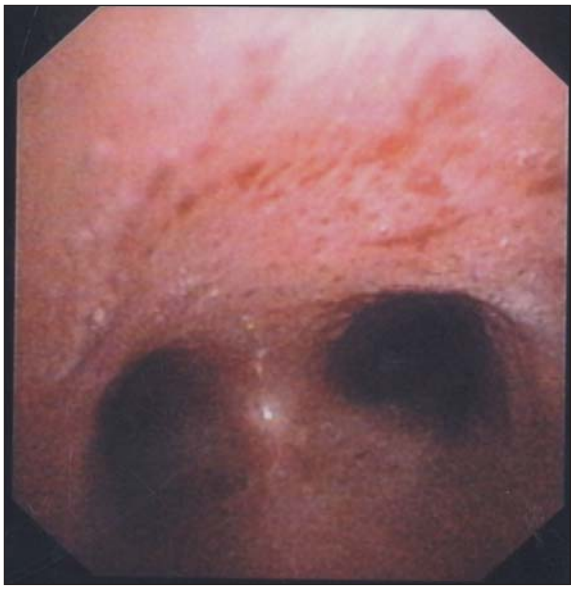

Figure 1. Purplish-red submucosal patches in the distal one-third of trachea.

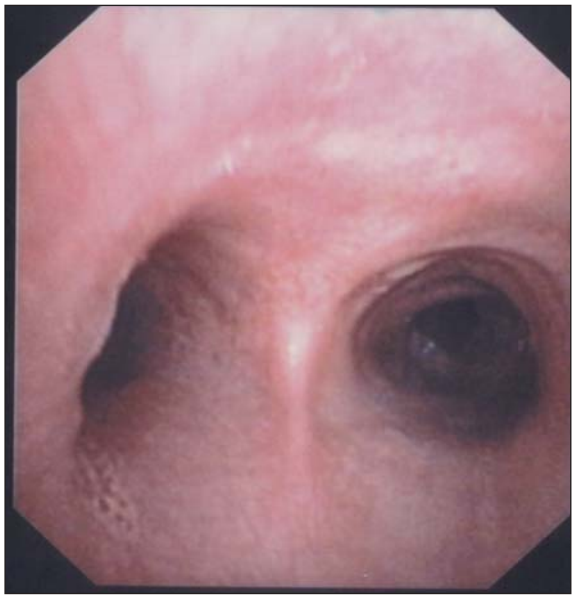

Figure 3. Follow-up bronchoscopic examination at the end of menstrual cycle showed disappearance of the previous distal one-third trachea.

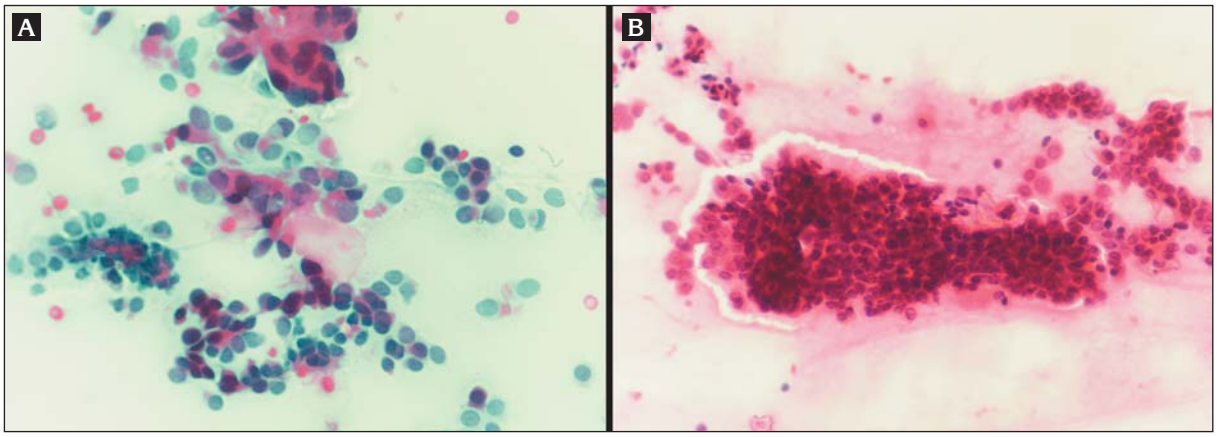

Figure 2. Photomicrograph of smear from bronchial brushing performed at the first day of the menstruation. Clusters of small cuboid cells consistent with endometrial stromal shedding; some bronchial ciliated epithelial cells are also demonstrated (A: Papanicolaou stain x400, B: Haemotoxylen-eosine stain x200).

Hemoptysis is a common clinical problem with various etiologies including catamenial hemoptysis (13). Experiencing hemoptysis in synchronal with menstrual periods by a female patient helps to differentiate catamenial hemoptysis from hemoptysis of other causes. The diagnosis of thoracic endometriosis is usually made on the basis of the clinical history and exclusion of other causes of recurrent hemoptysis (14). The bronchoscopic examination often yields inconclusive results due to distal parenchymal involvement rather than the mucosa of large bronchi (14-16). This may explain why histologic confirmation has been obtained in less than one-third of the previously reported cases, and why more invasive diagnostic procedures were required (9). Some investigators have suggested that when appropriate clinical and radiolographical findings are present, further tests including fiberoptic bronchoscopy are not indicated (16). Misdiagnosis is not rare because of the lack of prominent radiological findings, and it being a rather rare pathology. The present case was diagnosed two years after her first doctor visit.

It has been reported that thoracic endometriosis is either pleural (83%) or parenchimal (17%) (17). The pleura is the most commonly involved localization in thorasic endometriosis (17). Reports of catamenial hemoptysis suggesting intrapulmonary or bronchial involvement are less common (5). Tracheobronchial endometriosis is the least common form of the disease (13). We suggest that tracheobronchial endometriosis should be categorized as a distinct group of thoracic endometriosis due to its' difference from other subgroups of thoracic endometriosis with respect to clinical history, diagnostic role of bronchoscopy and treatment results with medical therapy (10). In the present case, almost whole tracheobronchial tree involving the distal trachea and bilateral main bronchia until the third branch of each lobar bronchi were involved.

Histological or cytological evidence to support diagnosis of pulmonary endometriosis is very important to avoid misdiagnosis or unnecessary drug therapy. Wang and colleagues reported four patients with tracheobronchial endometriosis. Cytologic features as well as the cyclic changes of the bronchoscopic findings in these cases were sufficient to diagnose tracheabronchial endometriosis. The diagnostic time intervals in their case series were significantly short because of prompt clinical suspicion and proper timing of bronchoscopic examination, which avoided unnecessary diagnostic procedures or the need for "doctor shopping", due to lack of definitive diagnosis. They also reported that the diagnostic role of computed tomography scan of the chest was not significant in their series. Similarly, in the present case diagnosis of endobronchial endometriosis was considered from the medical history. Further evaluations with bronchoscopic examination along with bronchial brushings from the suspected areas were performed. Although the patient spent two years without a diagnosis before her admission to our clinic, the diagnosis was made within eight days on the basis of a prompt suspicion followed by a diagnostic fiberoptic bronchoscopy.

The appropriate treatment for catamenial hemoptysis remains controversial due to the lack of large series in the literature. It is not clear whether to use medical treatment with hormones and hormone analogues or surgical removal of endometrial tissue. However, the medical therapy is generally recommended as the treatment of choice in pulmonary endometriosis (5). The aim of the medical treatment consists of suppression of ectopic thoracic endometrial tissue by using progesterone or danazol to cause pseudopregnancy or pseudomenopause (3-5). Danazol is a synthetic steroid with anti-estrogenic properties, which has proven to be effective in curing or controlling symptoms, even in patients who are not responsive to ovulation suppression (4,5). However, recurrence can occur when treatment is ceased. Danazol may also have severe side-effects, including climacteric symptoms such as virilisation, weight gain and sterility (4, 5). Some cases were successfully treated with GnRH agonist (18). GnRH agonist has few metabolic side effects, and the efficacy depends on the degree of ovarian suppression, which is related to the means of administration (18). Surgery is an option if the side effects of hormonal the-

rapy are intolerable, or if recurrence occurs when drug therapy is discontinued, or when the patient wishes to become pregnant (5). Endoscopic ablation is a new treatment option for catamenial hemoptysis; firstly, Puma and his colleaques reported a successfuly treated catamenial hemoptysis patient by endoscopic Nd-YAG laser ablation (2), and several years after Puma, Özvaran and his colleaques announced a new succesfuly treated catamenial hemoptysis patient by endoscopic argon laser ablation (8), and they proposed the laser treatment for the first line of therapy for central airway endometriosis without the adverse effects of pharmacologic therapy and surgical therapy (2,8). The present patient was treated medically with a GnRH analogue, and hormones including estrogen and progesterone in order to to prevent possible side effects of the GnRH analogue. She has not experienced another episode of hemoptysis under the above treatment during the last three months.

In conclusion, we suggest that, medical history with clinic suspicion, proper timing of bronchoscopic examination and cytologic evoluation of the bronchial brushing specimens are sufficient to warrant the diagnosis of tracheobronchial endometriosis, and to avoid misdiagnosis and unnecessary drug therapy.

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