

# A haemopneumothorax revealing thoracic endometriosis

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### ABSTRACT

Thoracic endometriosis is a rare disease but its actual frequency may be underestimated. Optimal management remains controversial. A multidisciplinary management approach, including surgery and hormone therapy, seems to give the best results and reduces recurrence. We report a case of a patient presenting with recurrent haemopneumothorax caused by thoracic endometriosis and treated surgically by video thoracoscopy and hormonal treatment.

**Key words:** pneumothorax, endometriosis, surgery, hormonal treatment

### Introduction

Endometriosis is a disease where tissue similar to the uterine lining grows outside the uterus.<sup>[1]</sup> This tissue has the same characteristics as the eutopic endometrium, thus responding to cyclical hormonal activity.<sup>[2,3,4]</sup> Different organs may be affected. There are four thoracic manifestations: catamenial pneumothorax (this is a spontaneous pneumothorax occurring within 72 hours after onset of menstruation), catamenial haemothorax, catamenial haemoptysis and pulmonary endometriotic nodules. Catamenial pneumothorax is the commonest form, most often located on the right side, characterized by diaphragmatic lesions (nodules, dehiscences) and by a particularly high recurrence rate.<sup>[5]</sup>

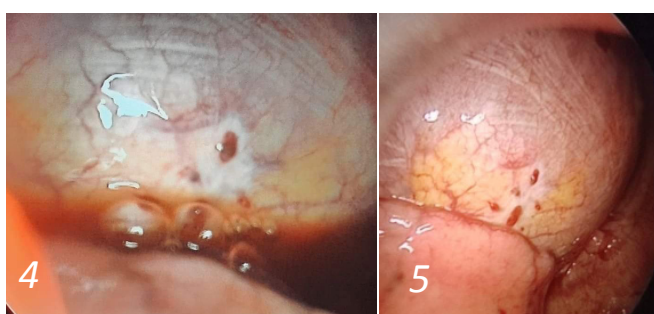
### Case Report

A 24-year-old, single lady, presented to the Department of Thoracic Surgery Algeria, with a third episode of haemopneumothorax five days after menstruation. Two years previously she had attended a pulmonology department with a dry cough, dyspnoea and chest pain. These symptoms were worse when supine and relieved by sitting up. Right chest drainage had been successful allowing the lung to re-expand. On her most recent presentation to our facility she had chest pain, dry cough and asthenia. Because of the recurrence of the effusion, she was referred to us for surgical pleurodesis. Chest radiography, ultrasound and chest CT scanning supported a mixed right hydropneumothorax. (Figures 1, 2 and 3). She underwent a video thoracoscopy which showed a haemothorax with multiple pleural and diaphragmatic nodules (Figures 4 and 5).

A resection of the diaphragmatic nodules was carried out with closure of the



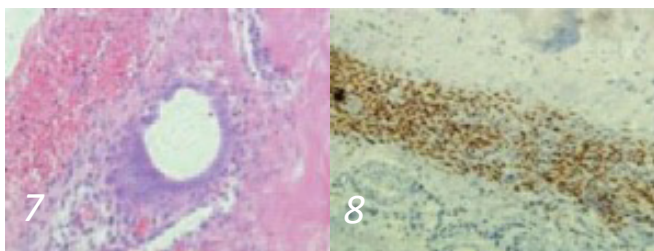
Figures 1, 2 and 3. Chest radiography, ultrasound and chest CT scanning supported a mixed right hydropneumothorax (Credit: Khalil Ghebouli Collection)



Figures 4 and 5. Videothoracoscopic appearance showing a haemothorax (Credit: Khalil Ghebouli Collection)



Figure 6. Videothoracoscopic appearance after pleurectomy (Credit: Khalil Ghebouli Collection)



Figures 7 and 8. Histological study of resection fragments diaphragmatic (Objective x20). Showing a localization of endometriosis. (Credit: Khalil Ghebouli Collection)

diaphragmatic breaches by direct suture associated with a parietal pleurectomy removing the pleural nodules (Figure 6).

The thoracic endometriosis was confirmed by histological study of the surgical specimen (Figures 7 and 8).

The patient was put on hormonal treatment based on danatrol/danazol: antigonadotropin) at 600 mg per day for six months. The short- and long-term evolution was without complications or recurrence. The patient was followed for two years.

### Discussion

Catamenial pneumothorax occurs in young women during the first 48 to 72 hours of the menstrual cycle. It was first described in 1958<sup>[6]</sup> and called “catamenial pneumothorax” by Lillington in 1972. Catamenial pneumothorax represents 2.8 to 5.6% of spontaneous<sup>[8]</sup> pneumothoraces in women, but may be an underestimate. It is not always easy to confirm this diagnosis, and it remains possible that a number of catamenial pneumothoraces are mistaken for primary spontaneous pneumothorax.

The mechanism of catamenial pneumothorax remains controversial. In the presence of pelvic endometriosis, catamenial pneumothorax must be actively sought. There are three hypotheses cited in the literature: anatomical, metastatic and physiological.

The anatomical hypothesis is based on the fact that during the menstrual cycle, the mucous plug at the cervix is absent and would allow the passage of air through the cervix and the tubes into the peritoneal cavity. This air would then migrate through diaphragmatic porosities in the thorax and thus create the pneumothorax.<sup>[6,7]</sup>

The metastatic hypothesis is also based on diaphragmatic fenestrations which might allow the passage of

endometriosis implants into the thoracic cavity via the lymphatic system. These implants, via the lymphatic chain or haematogenous route, may involve the pulmonary parenchyma, the visceral and/or parietal pleura, the diaphragm and rarely the tracheobronchial tract.

The physiological hypothesis states that a high serum level of prostaglandins F2 $\alpha$  during the menstrual cycle causes bronchospasms which may lead to alveolar ruptures causing pneumothorax. The rupture of emphysema bubbles is also more common during hormonal changes that occur during the menstrual cycle. The destruction during menstruation of ectopic endometrial tissue appears to cause rupture of the bubbles or blebs located in the visceral pleura.<sup>[10]</sup>

The cause of pneumothorax is likely to be multifactorial.

- It is exceptional to encounter a pneumothorax after laparoscopy, despite the presence of congenital diaphragmatic fenestrations.
- Recurrent catamenial pneumothorax has been described after hysterectomy.<sup>[11]</sup>
- Diaphragmatic fenestrations have been found in only 19 to 23% of catamenial pneumothoraxes.<sup>[11]</sup>
- Pelvic endometriosis is found in only 22 to 37% of cases of catamenial pneumothorax and thoracic endometriosis is found in only 23 to 35% of cases.<sup>[11]</sup>

However, in most cases, the likely mechanism is that of a transdiaphragmatic passage of air. The absence during the menstrual period of the cervical mucous plug could lead to the formation of a generally subclinical pneumoperitoneum and the passage into the thorax through diaphragmatic dehiscences, most often acquired secondary to diaphragmatic endometriosis.

Other manifestations of thoracic endometriosis, such as haemothorax and catamenial haemoptysis, are most often the consequence of bleeding linked to destruction of hypervascularized endometrial tissue.<sup>[11]</sup>

In rare cases, thoracic endometriosis manifests as isolated pulmonary nodules, without catamenial symptoms. These occur most often in older women, with less pronounced hormonal activity.<sup>[9]</sup>

The management of catamenial pneumothorax differs slightly from primary spontaneous pneumothorax. Video-assisted surgery makes it possible to identify and perform excision of endometriotic lesions which allow histological examination. Partial diaphragmatic resection is probably the

best treatment for lesions of the diaphragm (endometriotic nodules or perforations) so avoiding early recurrences which are reported after simple suture.<sup>[9]</sup> It also makes it possible to obtain tissue for detailed histological examination. Hormonal treatment interrupts the natural hormonal supply to endometrial tissue hence reducing proliferation of the endometriosis. danatrol (antigonadotropin) which causes atrophy of endometrial cells is frequently used at 600 to 800 mg per day for at least six months.<sup>[11]</sup>

Several side effects may occur such as weight gain, virilization and mood disorders. Triptorelin pamoate (Gn-RH analogue: gonadotropin-releasing hormone analogue like) can also be used at a dose of 3.75 mg per month (i.m.) for six to nine months.<sup>[11]</sup> However, hormonal treatments can cause permanent sterility limiting their use in women wanting to become pregnant and a surgical approach is favoured.

Surgery must precede medical treatment, allowing:

- Treatment of the cause of the pneumothorax (diaphragmatic perforation, visceral pleura implants, bullae, as appropriate).
- Resection of visible lesions with histological examination.
- Pleural symphysis by pleurectomy, pleural abrasion or talc instillation.

Hormonal treatment should be continued for 6 months and allows the pleura, diaphragm and lung to be "rested" from cyclical stresses for the period necessary for the definitive effectiveness of the symphysis.

## Conclusion

Thoracic endometriosis is an often undiagnosed cause of pneumothorax in women. The clinician must be alert to the possibility in women with recurrent, especially if right sided, features of pneumothorax. In most cases, diaphragmatic lesions are found and their excision allows treatment and histological examination. Multidisciplinary care is recommended.

Conflict of interest: None

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