

## Catamenial Pneumothorax : a Case Report and Review of the Literature

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**Key words.** Pneumothorax ; thoracoscopy ; endometriosis.

**Abstract.** Catamenial pneumothorax is a rare condition that is often misdiagnosed. It is defined as spontaneous pneumothorax occurring within 72 hours before or after onset of menstruation. Etiology is unknown but could be linked to endometriosis. Treatment is medicosurgical : thoracoscopy for pleural abrasion and hormone therapy to avoid recurrence.

### Introduction

Catamenial pneumothorax (CPT) is a rare entity of spontaneous, recurring pneumothorax in ovulating women (1). The etiology of this syndrome is unknown and many theories have been proposed (2). We report herein a case of catamenial pneumothorax and discuss medicosurgical treatment.

### Case report

A 37 year-old woman was admitted in emergency for a basic right-sided chest pain, without fever and no myocardial enzymes elevation. A chest X-ray showed a right pneumothorax without clinical nor radiological gravity signs (no tachypnea, no right ventricular insufficiency, no mediastinal shift, no bilateral pneumothorax). The patient had a previous history of two episodes of right pneumothorax in the last six months, always occurring during menstruation periods. The previous pneumothoraces were treated by simple exsufflation (Pleurocath®).

Considering the recurrence of the episodes, we decided to perform a video-thoracoscopy for abrasive mechanical pleurodesis. Surgical exploration revealed diaphragmatic fenestrations which were closed by simple closure but no endometriosis implant was found.

The post-operative course was marked by a recurrent right pneumothorax five days after ablation of the chest drainage. A new chest drainage was necessary for four more days. The patient was discharged after drain ablation without any complications.

According to the characteristics of this pneumothorax (spontaneous, recurrent, occurring in an ovulating woman), and despite the absence of pelvic endometriosis symptoms, we considered it as catamenial and started a hormonal therapy by a continuous progestative

treatment (lynestrenol) to stop menstruations and avoid recurrence.

Until the most recent follow-up (6 months), the patient is symptom-free and suffered no recurrence.

### Discussion

Catamenial pneumothorax is a rare entity (1) which is often underdiagnosed (2). It is defined as spontaneous pneumothorax occurring within 72 hours before or after onset of menstruation (3). Catamenial pneumothorax is, by definition, always correlated to the menstruation period. Nevertheless, not all menstruations lead to a catamenial pneumothorax in these patients.

The etiology of this syndrome is unknown (3). It is thought to involve pre-existing or acquired diaphragmatic defects and endometrial implants (4), although this latter condition is not always found (3, 4).

The main hypothesis to explain this syndrome is that the dissolving cervical mucous plug may allow the ascent of air through the fallopian tubes, causing a transient pneumoperitoneum (3). The intra-abdominal air subsequently escapes to the pleural space through the diaphragmatic defects, causing a catamenial pneumothorax.

MAUER and colleagues were first to associate CPT with endometriosis because they found erosive epiphrenic endometrial implants in their patients (5). Endometriosis affects 15% of all menstruating women mostly with pelvic manifestation (6). However, extrapelvic involvement has been encountered, including thoracic endometriosis (7). Catamenial pneumothorax is the most common manifestation of thoracic endometriosis (73%) (8). Nevertheless, pleural endometriosis is found in only half of the patients with catamenial pneumothorax, which were surgically explored (9, 10).

To explain endometriosis and its extent, three theories are relevant (2): retrograde menstruation and implantation, entry of endometrial cells into the venous system and spread of endometrial tissue and finally coelotomic metaplasia.

Catamenial pneumothorax is often located on the right side (2). This prevalence could be linked to the "piston effect" of the liver which transmits intraperitoneal pressure spikes across a perforated hemidiaphragm (11). Diaphragmatic endometrial implants are probably the cause of diaphragmatic defects (2). MARSHALL and colleagues (2) showed that all patients with diaphragmatic defects had endometrial implants. Other teams observed the presence of endometrial tissue at the borders of the defects (12).

Management of catamenial pneumothorax is a matter of controversy (2, 4). Some teams propose tubal ligation in case of recurrence (13). Because diaphragmatic defects are frequent and often occult, some teams propose a routine coverage of the diaphragmatic surface by a mesh to avoid recurrence (12, 14). Hormonal treatment in association with surgery is essential (2). Gonadotropin releasing hormone agonists (Gn-RH) are part of the pre or post operative management in high risk patients (2). Gn-RH agonists preserve the reproductive potential of the patient and are reversible treatments (15). They act by disrupting ovarian steroidogenesis (15).

In conclusion, one should suspect a catamenial pneumothorax in ovulating women presenting spontaneous pneumothorax, even in the absence of pelvic endometriosis symptoms (3). Treatment is medico-surgical: thoracoscopy for surgical inspection of the diaphragm (3) and surgical closure of all identified defects (3, 12, 14). Hormonal suppression is an important part of the treatment in conjunction with surgery to avoid recurrence (2, 14).

## References

1. OGER P, ALIFANO M., REGNARD J. F., GOMPEL A. Difficult management of recurrent catamenial pneumothorax. *Gynecol Endocrinol*, 2006, **22**: 713-15.
2. MARSHALL M. B., AHMED Z., KUCHARCZUK J. C. et al. Catamenial pneumothorax: optimal hormonal and surgical management. *Eur J Cardiothorac Surg*, 2005, **27**: 662-66.
3. KOROM S., CANYURT H., MISSBACH A. et al. Catamenial pneumothorax revisited: clinical approach and systematic review of the literature. *J Thorac Cardiovasc Surg*, 2004, **128**: 502-8.
4. PEIKERT T., GILLEPSIE D. J., CASSIVI S. D. Catamenial pneumothorax. *Mayo Clin Proc*, 2005, **80**: 677-80.
5. MAURER E. R., SCHAAL J. A., MENDEZ F. L. Jr. Chronic recurring pneumothorax due to endometriosis of the diaphragm. *JAMA*, 1958, **168**: 2013-14.
6. CANDIANI G. B., VERCELLINI P., FEDELE L. et al. Mild endometriosis and infertility: a critical review of epidemiologic data, diagnostic pitfalls, and classification limits. *Obstet Gynecol Surv*, 1991, **46**: 374-82.
7. KARPEL J. P., APPEL D., MERAV A. Pulmonary endometriosis. *Lung*, 1985, **163**: 151-59.
8. JOSEPH J., SAHN S. A. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. *Am J Med*, 1996, **100**: 164-70.
9. SHIRAIISHI T. Catamenial pneumothorax: report of a case and review of the Japanese and non-Japanese literature. *Thorac Cardiovasc Surg*, 1991, **39**: 304-7.
10. SCHOENFELD A., ZIV E., ZEELEL Y. et al. Catamenial pneumothorax: a literature review and report of an unusual case. *Obstet Gynecol Surg*, 1986, **41**: 20-4.
11. KIRSCHNER P. A. Porous diaphragm syndromes. *Chest Surg Clin N Am*, 1998, **8**: 449-72.
12. BAGAN P., LE PIMPEC BARTHES F., ASSOUD J. et al. Catamenial pneumothorax: retrospective study of surgical treatment. *Ann Thorac Surg*, 2003, **75**: 378-81.
13. LAURSEN L., ØSTERGAARD A. H., ANDERSEN B. Catamenial pneumothorax treated by laparoscopic tubal occlusion using Filshie clips. *Acta Obstet Gynecol Scand*, 2003, **82**: 488-89.
14. LEONG A. C., COONAR A. S., LANG-LAZDUNSKI L. Catamenial pneumothorax: surgical repair of the diaphragm and hormone treatment. *Ann R Coll Surg Engl*, 2006, **88**: 547-49.
15. AKAL M., KARA M. Nonsurgical treatment of a catamenial pneumothorax with a Gn-RH analogue. *Respiration*, 2002, **69**: 275-76.