

# MENSTRUAL CYCLE RELATED PNEUMOTHORAX: CASE REPORT AND REVIEW OF THE LITERATURE

Nika Oreskovic<sup>1</sup>, Željko Djaković<sup>1</sup>, Ivka Djakovic<sup>2</sup>, Krunoslav Kuna<sup>2</sup>

## ABSTRACT

**Background:** Catamenial pneumothorax is the most common form of thoracic endometriosis syndrome. It occurs around the beginning of a menstrual cycle. Although the mechanism of catamenial pneumothorax is not definitely clear, endometriosis plays an important role in it. Video-assisted thoracic surgery is standard procedure for the treatment of recurrent pneumothorax in general.

**Case study:** We report on a case of catamenial pneumothorax in women with a history of recurring spontaneous pneumothoraces associated with diaphragmatic endometrial implants who is involved in the IVF procedure.

**Conclusion:** Combination of video-assisted thoracoscopic surgery (VATS) and gonadotropin-releasing-hormone analogue gives the best results, to reduce the risk of pneumothorax to recur. Treatment of catamenial pneumothorax is complex and should include thoracic surgeon and gynecologist as soon as the diagnosis is definitive.

## Keywords:

catamenial pneumothorax, video-assisted thoracic surgery, pelvic endometriosis, thoracic syndrome

## INTRODUCTION

Catamenial pneumothorax (CP), derived from the Greek word "katamenios" which suggests its monthly occurrence, is a clinical condition defined as pneumothorax occurring within 72 hours of menstruation [1,2]. According to Mehta *et al*, CP is one of the most underdiagnosed etiological feature of primary spontaneous pneumothorax (PSP) found in young healthy menstruating women who addressed more than one case of clinical manifestation of dyspnoea and chest pain followed by or in coexistence with menses [3]. Although underdiagnosed, only 3% to 6% of spontaneous pneumothorax cases meet the definition of catamenial pneumothorax, establishing an indication for surgical treatment in only as 1/3 of the affected women as it was stated by Barbosa *et al*. [4].

Generally, CP was considered a rare phenomenon in history of its tracking with a previously reported

prevalence of 0.9 – 5.6% of all cases of PSP in women, but with the progress in radiological diagnostics and better understanding of the condition, the awareness has been raised and the number of diagnosed cases has grown to 18 -30% [1,4].

The mean age of onset is 32-35 years in ovulatory women, while the women under hormonal therapy (such as contraceptive medication), the pregnant ones and women in menarche are generally not subject to it [1,3,5,6]. The localization of lesion found according to a review by Takahashi *et al*. is predominantly on the right side of the chest (85-95%), even though it may also occur on the left side (4.8%) or bilaterally (3.5%) [7].

There are three main criteria that determine the diagnosis of CP. Along with previously referred occurrence of the pneumothorax meticulously linked to ongoing menstruation, two subsequent criteria are clarified as: absence of any primary lung disease where pneumothoraces initiated secondary to a known underlying lung disease has not been classified as catamenial and at least one recurrent event (minimum of two episodes of pneumothorax) to achieve the definition criteria. [3-5,8].

While reevaluating the diagnosis of the CP before the surgical treatment, chest radiography, less often computed tomography (CT), and rarely magnetic resonance imaging (MRI) are performed. The fact radiography is routinely performed; there are no specific imaging diagnostic criteria [3,9].

When choosing the best treatment method for catamenial pneumothorax, several techniques have been proposed in literature. Nonetheless, video-assisted thoracoscopic surgery (VATS) has found to be the treatment of choice [1-10]. If possible, it is advisable to perform VATS during menstruation, because it allows maximum visibility of the potential endometriotic implants [9].

Thoracotomy is indicated almost exclusively in cases of recurrence after a previous procedure [11]. The use of video-assisted mini-thoracotomy (VAMT) has been suggested if the procedure involves extensive lesions within the diaphragm [8].

<sup>1</sup>Department of Emergency Medicine, Insititution for Emergency Medicine of Zagrebačka County

<sup>2</sup>Clinical Department of Gynecology and Obstetrics, University Hospital Centre Sestre milosrdnice, Zagreb, Croatia

**Corresponding author:** Nika Oreskovic, Department of Emergency Medicine, Insititution for Emergency Medicine of Zagrebačka County, Croatia

**e-mail:** oresko.nika@gmail.com

**DOI:** 10.5281/zenodo.4010887

## CASE STUDY

A 33-year-old woman was admitted at thoracic surgery department for second onset of right pneumothorax in two months. After evaluation video-assisted thoracoscopic surgery (VATS) procedure was indicated and performed in a standard mode. Catamenial pneumothorax was suspected by clinical signs. Elective thoracic surgery procedures are not conducted during menstruation, so we operated after cessation of menstrual bleeding. The diagnosis of catamenial pneumothorax is rarely confirmed by histopathology as in our case. Three port VATS was performed, apical segment of the upper lobe resected and pleurodesis achieved by termocoagulation of the upper half of parietal pleura. During procedure typical lesions of the diaphragm were found (Figure 1). Additional investigation revealed that both onsets of pneumothorax occurred on first day of menstrual bleeding. CA 125 level was 49.9 kIU/L (normal <35 kIU/L). Ultrasonography (US) examination showed a typical endometrial cystic formation measuring 25 mm on the right ovary. Gonadotropin-releasing-hormone analogue therapy was recommended but patient was trying to get pregnant after a series of miscarriage and rejected therapy. Two months after VATS procedure she had symptoms of pneumothorax and x-ray revealed partial subpulmonal pneumothorax that did not required treatment (Figure 2). Positron emission tomography scan was performed and suspected endometriosis at the abdominal side of the diaphragm was found. In the follow up period she had one more episode of pneumothorax like symptoms at the beginning of menstrual bleeding. Patient is currently in the *in vitro* fertilization procedure.

## DISCUSSION

We report on a case of catamenial pneumothorax in women with a history of recurring spontaneous pneumothoraces associated with diaphragmatic endometrial implants who is involved in the *IVF* procedure.

Given the fact that pneumothorax is reported in concordance with menstruation, followed by the coexisting endometrial extrauterine tissue findings, suggests the presence of an undeniable connection between these two entities [1-12]. First association of CP with endometriosis was acknowledged by the Maurer *et al.*, who found erosive epiphrenic implants in patients suffering from recurrent spontaneous pneumothoraces whose appearance coincide with the beginning of menstruation [8]. Endometriosis, described as any endometrial tissue found outside of the uterine cavity, affects 5% to 15% of women in reproductive age [1-5,7-13]. Although it appears to be one of the most common benign gynecological proliferations, this disease remains poorly understood. Studies reviewed by Mehedintu *et al.* implicate there is

no relationship between the extent of the disease and its symptomatology [14].

According to the previously mentioned, it is not the quantity of endometrial foci rather the localization that results with recurring pneumothorax. Several theories concerning the endometriosis related CP has been proposed. They all conflate around the idea of increased fallopian tube permeability in peri menstrual period combined with the fenestration in diaphragmic wall due to a congenital defect or more often seen endometrial metastatic lesions found on the surface of diaphragm that can damage the soft tissue of diaphragm and open the pathway to lung pleura [1,7,8,14-17]. In some cases the endometrial cells continue their migration through the diaphragm and form endometrial nodules on the surface of the pleura [8]. The reason why women's period represents the inevitable risk factor for a threatening pneumothorax lies in the fact that endometrial tissue attached to the pleura becomes physiologically active [7-13]. Influenced by gonadotropins released during menstruation, endometrial cells free the prostaglandin molecules which then corrode the small pulmonal vessel and alveolar septa culminating with occurrence of pneumothorax [18]. Accompanied by hematochezia, hemothorax and radiologically apparent endometrial nodules on the pleura it refers to a specific entity called thoracic endometriosis syndrome (TES). It's occurrence correlates with existence of pelvic endometriosis in around 50 – 70%, where the thoracic endometriosis occurred approximately 5 years later [3,7,14-18]. As previously detailed, difference in the mean age occurrence between pelvic and thoracic endometriosis may be explained with the time necessary for endometrial tissue to migrate through the right diaphragm [16].

Catamenial pneumothorax is found to be associated with both thoracic and pelvic endometriosis, although endometrial character of the disease cannot be confirmed histologically in every case [7,9]. From the clinical point of view, CP displayed to be leading cause of primary spontaneous recurrent pneumothorax with prevalence of almost 30% in emergency room [1-6]. When reviewing the literature upon the data on statistical correlation between catamenial pneumothorax and underlying endometriosis, the findings showed to be following.

From the prospective research of Rousset- Jablonski *et al.* where 156 women were surgically treated for spontaneous pneumothorax, we discovered that histologically documented thoracic endometriosis was found in 23.1% (36/156) of all patients (including catamenial and non-CP patients), being 6.5 times more frequent in patients with catamenial pneumothorax. As expected, pelvic endometriosis was found in about 50% of thoracic endometriosis cases, with wide variation among studies suggesting even higher rate of occurrence. In the analysis of 110 thoracic endometriosis cases by Joseph *et al.*, pelvic endometriosis was found

in 84% (51/61) of investigated patients with thoracic endometriosis [1].

The overall small number of reported cases suffering from thoracic endometriosis comes from the fact that not only it is a rare condition; endometriosis in the visceral pleura might be overlooked during surgery because endometrial tissue disappears at certain points during the menstrual cycle [7].

When investigating the possible causes of PSP during the first encounter with the patient, it is of crucial importance not only to ask for the last period commencement then to search for the pelvic pain and infertility history where pelvic exam including pelvic imaging is recommended [14,16-20]. In addition to previously mentioned, of great diagnostic value is the measurement of cancer antigen (CA)125 in cases of CP. As observed in research of Tsunezuka *et al.* in the female patients diagnosed with endometriosis related pneumothorax after VATS procedure, serum CA125 level was very high before the surgery compared to the control group of disease-free females [21].

As illustrated in this case report, our patient has had a history of miscarriage and was in the process of in vitro fertilization during hospitalization. After she presented with pneumothorax we opted for VATS, the video assisted thoracic surgery, where the suspected endometrial tissue was found on the inner wall of the diaphragm. Despite the fact that the procedure has been successfully carried out several episodes of reoccurring pneumothorax were documented. Their reoccurrence originates due to rejected analogues hormonal therapy recommended for lowering the recurrence risk [22].

The foundation of treatment for endometriosis-related pneumothorax, even before undergoing the VATS procedure, is the suppression of ovarian oestrogen secretion. In the long history of CP management commonly applied medications were oral contraceptives, progesterone agents, danazol or gonadotropin-releasing hormone (GnRH) agonists [23-25]. Although the preceding two have been used most frequently they all share similar mechanism. Women's self-regulated GnRH receptors are suppressed with GnRH analogues causing down-regulation and creating reversible hypogonadotropic hypogonadism. In previous years, hormone therapy has been the backbone treatment of CP. However through years, VATS was introduced as a method of choice and hormone therapy took place as maintenance therapy preventing the recurrence of the pneumothorax. The reason for that lays in the fact that hormonal therapy needed to be maintained for prolonged period of 6 months causing loss of trabecular bone density, hot flashes and vaginal dryness which presented as large drawback for its utilization [25-27].

In regard with our patient's wishes to conceive, estrogen suppression was contraindicated.

## CONCLUSION

Leakage of air in pleural space due to the rupture of alveolar septa influenced by women's menstrual cycle, which is shown to be recurrent in its nature, is defined as catamenial pneumothorax. Because of the high recurrence rate despite the therapy, a need evolved to determine the underlying cause to make the treatment more effective and to lower the recurrence to a minimum. Combination of VATS and gonadotropin-releasing-hormone analogue gives the best results, to reduce the risk of pneumothorax to recur. Treatment of catamenial pneumothorax is complex and should include thoracic surgeon and gynecologist as soon as the diagnosis is definitive.

## CONFLICT OF INTEREST:

**The authors declare that there is no conflict of interest.**

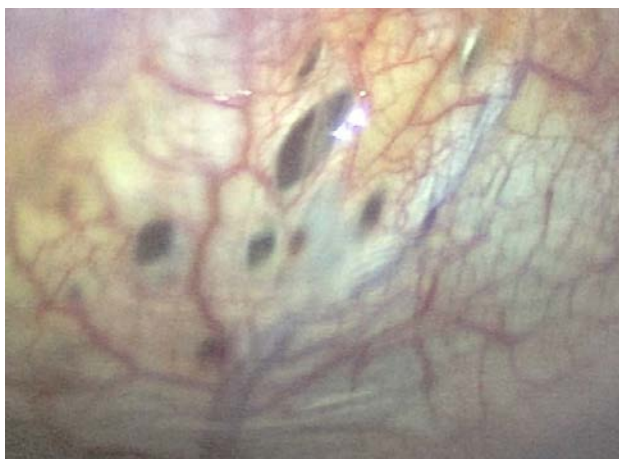
**The patient gave her informed consent prior to her inclusion in case report.**

## REFERENCES

1. Aissa S, Benzarti W, Alimi F, Gargouri I, Salem HB, Aissa A *et al.* Catamenial pneumothorax revealing diaphragmatic endometriosis: a case report and revue of literature. *Pan Afr Med J.* 2017 14; 27: 112.
2. Marjański T, Sowa K, Czapl A and Rzyman W. Catamenial pneumothorax – a review of the literature. *Kardiochir Torakochirurgia Pol.* 2016 13(2): 117–121.
3. Mehta CK, Stanifer BP, Fore-Kosterski S, Gillespie C, Yeldandi A, Meyerson S *et al.* Primary Spontaneous Pneumothorax in Menstruating Women Has High Recurrence. *Ann Thorac Surg.* 2016;102(4):1125-30.
4. Barbosa BC, Marchiori E, Zanetti GMR, and Barillo JG. Catamenial pneumothorax. *Radiol Bras.* 2015; 48(2): 128–9.
5. Visouli A, Zarogoulidis K, Kougoumtzi I, Huang H, Li O, Dryllis G *et al.* Catamenial pneumothorax. *J Thorac Dis.* 2014; 6(Suppl 4): S448–S460.
6. Kolos A, Dzhieshev Z, Dikolaev V, Amangaliev A. Catamenial Pneumothorax- poster presentation. *Exp Clin Transplant.* 2015;13 Suppl 3:144-5.
7. Takahashi R, Kurihara M, Mizobuchi T, Ebana H and Yamanaka S. Left-Sided Catamenial Pneumothorax with Thoracic Endometriosis and Bullae in the Alveolar Wall. *Ann Thorac Cardiovasc Surg.* 2017; 23(2): 108–112.
8. Fukuoka M, Kurihara M, Haga T, Ebana H, Kataoka H, Mizobuchi T. Clinical characteristics of catamenial and non-catamenial thoracic endometriosis related pneumothorax. *Respirology* 2015;20(8):1272-6
9. Korom S, Canyurt H, Missbach A, Schneiter D, Kurrer MO, Haller U. Catamenial pneumothorax revisited: clinical approach and systematic review of the literature *J Thorac Cardiovasc Surg.* 2004;128(4):502-8
10. Rousset-Jablonski C, Alifano M, Plu-Bureau G, Camilleri-Broet S, Rousset P, Regnard JF *et al.* Catamenial pneumothorax and endometriosis-related pneumothorax: clinical features and risk factors. *Human Reproduction.* Vol:26, No:9, 2011.
11. Ouede R, Alexandre BD, Gregoire AK, Kohou-Kone L, N'guessan E, Kouacou MG *et al.* Pneumothorax catamenial: results of 18 cases operas. *Pan Afr Med J.* 2018 25;30:168.
12. Ottolina J, De Stefano F, Viganò P, Ciriaco P, Zannini P, Candiani M. Thoracic Endometriosis Syndrome: Association With Pelvic Endometriosis and Fertility Status. *J Minim Invasive Gynecol.* 2017;24(3):461-5.

13. Bricelj K, Srpčič M, Ražem A, Snoj Ž. Catamenial pneumothorax since introduction of video-assisted thoracoscopic surgery: A systematic review. *Wien Klin Wochenschr.* 2017;129(19-20):717-26.
14. Mehedintu C, Plotogea MN, Ionescu S, Antonovici M. Endometriosis still a challenge. *J Med Life.* 2014 15;7(3):349-57.
15. Shikino K, Ohira Y, Ikusaka M. Catamenial pneumothorax. *J Gen Intern Med.* 2016;31(10):1260.
16. Alifano M, Roth T, Broët SC, Schussler O, Magdeleinat P, Regnard JF. Catamenial pneumothorax: a prospective study. *Chest.* 2003;124(3):1004-8.
17. Goorsenberg AWM, Pruis M, Boshuizen RC, Hindori V, Slaar A, Bresser P. Catameniale pneumothorax [Catamenial pneumothorax: an intriguing cause of recurrent pneumothorax in women]. *Ned Tijdschr Geneesk.* 2018;162:D2340.
18. Tulandi T, Sirois C, Sabban H, Cohen A, Murji A, Singh SS et al. Relationship between Catamenial Pneumothorax or Non-catamenial Pneumothorax and Endometriosis. *J Minim Invasive Gynecol.* 2018;25(3):480-3.
19. Attaran S, Bille A, Karenovics W, et al. Videothoracoscopic repair of diaphragm and pleurectomy/abrasion in patients with catamenial pneumothorax: a 9-year experience. *Chest.* 2013;143:1066-9.
20. Baisi A, Raveglia F, De Simone M, Cioffi U. Catamenial Pneumothorax: A Matter of Anamnesis. *Ann Thorac Surg.* 2017;104(1):367-68.
21. Bagan P, Berna P, Assouad J, et al. Value of cancer antigen 125 for diagnosis of pleural endometriosis in females with recurrent pneumothorax. *Eur Respir J.* 2008;31:140-2.
22. Costa F, Matos F. Endometriose torácica. *Rev Port Pneumol.* 2008;XIV:427-35.
23. Cassina PC, Hauser M, Kacel G, et al. Catamenial hemoptysis. Diagnosis with MRI. *Chest.* 1997;111:1447-50.
24. Garner M, Ahmed E, Gatiss S, West D. Hormonal manipulation after surgery for catamenial pneumothorax. *Interact Cardiovasc Thorac Surg.* 2018 1;26(2):319-322.
25. Inam H, Inam S, Tahir M. Catamenial pneumothorax: A case report. *J Pak Med Assoc.* 2016;66(10):1327-1329.
26. Lalani S, Black A, Hodge MC, Tulandi T, Chen I. Dienogest Therapy as a Treatment for Catamenial Pneumothorax: Case Report and Review of Hormonal Options. *J Obstet Gynaecol Can.* 2017;39(9):764-768.

## FIGURES



**Figure 1.** Intraoperative finding of diaphragmal lesion.



**Figure 2.** Chest x-ray shows subpulmonal pneumothorax.