

# Endometriosis - A rare cause of primary spontaneous pneumothorax

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## SUMMARY

**Catamenial pneumothorax is a rare condition. We report a case of a 36-year-old female who presented with dyspnoea every time before she had her regular menses. Further investigation confirmed that she had catamenial pneumothorax. With this case we wish to highlight this rare diagnostic entity that every clinician should keep in mind.**

## INTRODUCTION

Endometriosis is defined as the presence of ectopic endometrial tissue outside the uterine cavity.<sup>1</sup> Endometriosis commonly involves the pelvis, and it is rare that endometriotic tissues can be found outside the pelvis in the abdomen, thorax, brain or skin.<sup>1,2</sup> Thoracic involvement is the most frequent extra-pelvic location of endometriosis.<sup>1</sup>

## CASE REPORT

We would like to report a rare case of catamenial pneumothorax. A 36-year-old female experienced mild dyspnoea and right sided chest pain for two to three days before the start of her regular menses for the last three years. The dyspnoea and chest pain normally improved once she starts to have her menses. Her menstrual cycles were regular with menses lasting around 5-7 days and each cycle would last around 28-30 days. She denied taking any form of oral contraception pills and had no history of endometriosis. Her first chest x-ray (CXR) done showed a 1cm right pneumothorax. The rim of pneumothorax was not increasing in size on serial chest x-ray and hence, no intervention was done initially. She was discharged. However, after two weeks her dyspnoea worsened, and repeated chest x-ray showed worsening pneumothorax and a chest tube was inserted. She had persistent air leak despite prolonged drainage and hence was referred to the cardiothoracic surgeon for a video-assisted thoracoscopic surgery (VATS) to examine her pleural cavity. During the VATS procedure, pigmented deposits were seen at both the parietal and visceral pleural layers (Figure 1). Biopsy taken confirmed the presence of the endometriotic implants. Surgical pleurodesis was done during the VATS procedure. Four months after the VATS procedure, unfortunately she developed a right tension pneumothorax requiring a chest drain. Her computed tomography of thorax showed right moderate pneumothorax with no evidence of

pneumomediastinum, bullae, pleural nodules, cysts, cavity or ground glass infiltrates. She adamantly refused a second surgical referral for her recurrent pneumothorax. Hence, when her lungs re-expanded after drainage of 6 days, chemical pleurodesis was attempted. She was given bleomycin 60mg into her right thoracic cavity. Her first bleomycin pleurodesis failed and her pneumothorax recurred two hours post procedure. She was subjected to further drainage for about four days before a second attempt of chemical pleurodesis was made when the lungs re-expanded again. This time autologous blood patch was attempted, and it was successful. Talc pleurodesis was not available during that period. The gynecology team started her on monthly intramuscular leuprolide (Lucrin) 3.75mg injection for 6 months and subsequently dienogest which suppresses ovulation and serum oestrogen levels along with barrier contraception. During her follow-up for the last one year, she remained asymptomatic. Prior to the event she had had two children.

## DISCUSSION

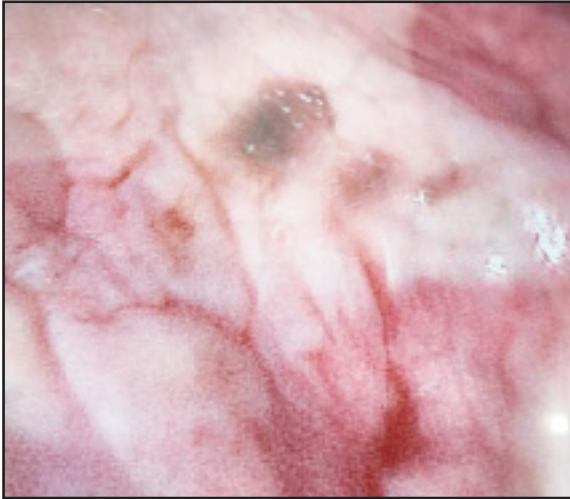
Catamenial pneumothorax is a rare entity which is often under diagnosed.<sup>1</sup> It is defined as recurrent spontaneous pneumothorax occurring within the window of 24 hours before to 72 hours after the onset of menses.<sup>1,2</sup> Thoracic endometrial implants can then cause catamenial pneumothorax by several mechanisms. The ectopic endometriotic implant can result in spontaneous rupture of blebs, leading to pneumothorax.<sup>2,3</sup> It causes alveolar rupture mediated by prostaglandin-induced bronchiolar constriction. The sloughing of endometrial implants of visceral pleura may result in air leaks. Passage of air from the genital tract through diaphragmatic defects in the absence of the cervical mucous plug during menses may also lead to episode of catamenial pneumothorax.<sup>2,3</sup>

The most common presentation of thoracic endometriotic syndrome is catamenial pneumothorax (73%), while catamenial haemothorax is present in just 14% of the cases, followed by haemoptysis (7%) and lung nodule (6%).<sup>3</sup> Reports in the literatures describe a common right sided involvement. However, clinician should not exclude the diagnosis of thoracic endometriosis in a patient with left sided thoracic involvement. Naureen Narula et al., reported a rare clinical entity of left sided thoracic endometriosis.<sup>2</sup>

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**Fig. 1:** During VATS procedure, endometrial implants (raised pigmented lesions) were seen on her right pleural and diaphragmatic surfaces with size ranging from 0.5 cm<sup>2</sup> to 2 cm<sup>2</sup>.



**Fig. 2:** Computed tomography of the thorax showed right moderate pneumothorax with chest tube in-situ and partial collapsed of the right lung.

Surgical intervention should be the first line of correction for possible leak of a recurrent pneumothorax. The treatment of catamenial pneumothorax can be divided into hormonal and surgical therapy.<sup>4</sup> Hormonal therapy aims at blocking the hormonal support from ovary by inducing a pseudopregnancy or pseudomenopausal state.<sup>4,5</sup> Commonly used agents are oral contraceptives, progesterone agents, danazol or gonadotrophin-releasing hormone analogs (GnRH).<sup>4</sup> In the recent years, GnRH analogs have been advocated as first line therapy due to its effectiveness.<sup>5</sup> However, its use is limited by the side effects of menopause and risk of osteoporosis. Unfortunately, treatment with hormonal therapy alone is associated with a recurrence rate exceeding 50% within six months after treatment was stopped.<sup>5</sup>

#### CONCLUSION

Although catamenial pneumothorax is a rare disease, it should be suspected in females of reproductive age group who present with recurrent spontaneous pneumothorax.

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