

CASE REPORT

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Catamenial pneumothorax and endometriosis: Report of a rare case

Oluwaseun Adelekan, Moaz Alowami, Pooja Vasudev

ABSTRACT

Introduction: Catamenial pneumothorax is a rare condition which is characterized by the recurrent accumulation of air within the pleural cavity without related respiratory diseases. It happens in women of reproductive age, usually within 72 hours before or after onset of menstruation. It occurs in association with characteristic pleural lesions, right-sided location of the pneumothorax, and concomitant thoracic endometriosis. The objective is to report a rare case of thoracic endometriosis in a 35-year-old woman with recurrent catamenial pneumothorax.

Case Report: A 35-year-old female presented with persistent and recurrent right pneumothorax usually occurring immediately after her menstruation. She had no personal history of lung problems, asthma, bronchitis, or any family history of any chronic lung disease. Computed tomography (CT) of the chest showed a large hydropneumothorax on the right side resulting in near-complete collapse of the right lung and mediastinal shift from right to left. The gross specimen comprised the right upper lobe resection of the lung, which weighed 4.9 grams and measured 7.7 cm × 1.5 cm × 1.2 cm. The pleura was mottled brown-gray with adhesions. Histopathologic findings showed pleural endometriosis (pleural endometrial glands and stroma present), consistent with catamenial pneumothorax. Associated pleural

adhesions with acute and chronic inflammation and mild emphysematous changes in the lung parenchyma were also noted. Immunohistochemistry with immunostains estrogen receptor (ER) and CD10 highlighted the endometrial glands and stroma which confirmed the diagnosis.

Conclusion: There should be a high index of suspicion of catamenial pneumothorax in women of reproductive age who present with pneumothorax during the perimenstrual period. Adequate sampling of lung wedge resections and careful microscopic examination, followed by confirmation with immunohistochemistry, play important roles in the diagnosis of this rare condition.

Keywords: Catamenial pneumothorax, Endometriosis, Immunohistochemistry

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Oluwaseun Adelekan¹, Moaz Alowami², Pooja Vasudev¹

Affiliations: ¹Department of Pathology and Molecular Medicine, McMaster University, St Joseph's Healthcare L222-5, St Luke Wing, 50 Charlton Avenue East, Hamilton, Ontario L8N 4A6, Canada; ²Department of Radiology, St Joseph's Healthcare, 50 Charlton Avenue East, Hamilton, Ontario L8N 4A6, Canada.

Corresponding Author: Oluwaseun Adelekan, Department of Pathology and Molecular Medicine, McMaster University, St Joseph's Healthcare L222-5, St Luke Wing, 50 Charlton Avenue East, Hamilton, Ontario L8N 4A6, Canada; Email: Oluwaseun.adelekan@medportal.ca

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INTRODUCTION

Catamenial pneumothorax (CP) was first described by Lillington et al. in 1972 [1]. It is considered as a rare condition and subtype of pneumothorax which is related to menstruation and endometriosis. This condition is usually misdiagnosed and characterized by the recurrent accumulation of air within the pleural cavity without concomitant respiratory diseases. The typical clinical manifestation is spontaneous pneumothorax, or this condition may also be asymptomatic. It happens in reproductive age women, usually within 72 hours of onset of menstruation [2]. Thoracic endometriosis, right side involvement, and distinctive pleural lesions (multiple

diaphragmatic fenestrations, brown nodules on the diaphragm) are characteristic of this condition [2].

CASE REPORT

A 35-year-old woman, premenopausal, para 1 presented to the emergency department with sudden onset of shortness of breath and chest pain. This episode coincided with the first three days of her menstruation. She alluded to prior episodes of left shoulder pain and chest pain occurring during her menstruation. She also had a past history of a spontaneous right hydropneumothorax four weeks prior to presentation for which she had a chest tube insertion drainage in the emergency room as well as care in the intensive care unit. Additionally, there was also a history of a small left apical pneumothorax two years prior to presentation which required no intervention. A history of pelvic and peritoneal endometriosis, infertility and 2 laparoscopies of the pelvis was also noted. However, there was no history of chronic lung disease, recent surgical procedures, or trauma. There was no additional pertinent personal or family medical history.

Clinical examination revealed tachypnea, mediastinal shift from right to left and reduced breath sounds. The laboratory results were within reference range. Chest X-ray showed right-sided moderate to large hydropneumothorax (Figure 1). A chest CT performed, revealed bilateral pleural nodularity in keeping with pleural-based endometriosis deposits and right basal atelectatic changes also noted (Figure 2). A transvaginal ultrasound (US) showed severe endometriosis including extra-pelvic endometriosis of the abdominal wall. Specifically, rectal and severe posterior compartment endometriosis was noted. There was also severe obliteration of the rectouterine pouch and benign cystic lesion in the left adnexa. There was free blood in the right adnexa likely due to a retrograde menstruation. In spite of several chest drains, her lung did not expand and she continued to have persistent air leaks. She underwent a flexible video-assisted bronchoscopy, right video-assisted thoracoscopic surgery (VATS). Intra-operatively, there were adhesions seen between the right lung and the chest wall and an area of abnormality on the lateral wall of the right upper lobe between the apical and posterior segments. The pleura was also abnormal in this area. A wedge resection of the right upper lobe of the lung was performed and the specimen was sent for pathology analysis.

Grossly, the specimen comprised a wedge resection of the upper lobe of the right lung, which measured 7.7 cm × 1.5 cm × 1.2 cm and weighed 4.9 grams. The pleura of the lung was mottled brown-gray with adhesions. The lung parenchyma was tan and appeared relatively unremarkable. Microscopic examination showed endometrial glands and stroma surrounded by hemorrhage as well as fibrosis of the pleura (Figure 3). There was also acute and chronic inflammation in addition

to subpleural chronic inflammation (Figure 4). Mild emphysematous change of lung parenchyma was also noted. Immunohistochemical stains performed confirmed the endometrial glands and stroma to be estrogen receptor (ER) positive (Figure 5) and the endometrial stroma to be CD10 positive (Figure 6).

The postoperative course was uneventful, and the patient was discharged four days after surgery. She was subsequently placed on Leuprolide injections every three months and Norlutate as an add-back hormone for a period of one year. She has not had any recurrence of shortness of breath or chest pain in the last two years following her surgery. She remains quite stable from a thoracic standpoint. However, she continues to experience pelvic and abdominal pain while on medical management and is scheduled to have a laparoscopic resection of bowel endometriosis.

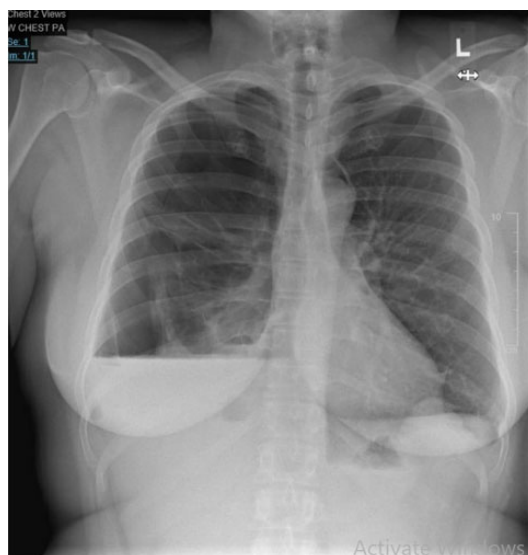


Figure 1: Chest X-ray demonstrating a right-sided moderate to large hydropneumothorax.



Figure 2: Chest CT scan showing bilateral pleural nodularity in keeping with pleural-based endometriosis deposits and right basal atelectatic changes also noted.

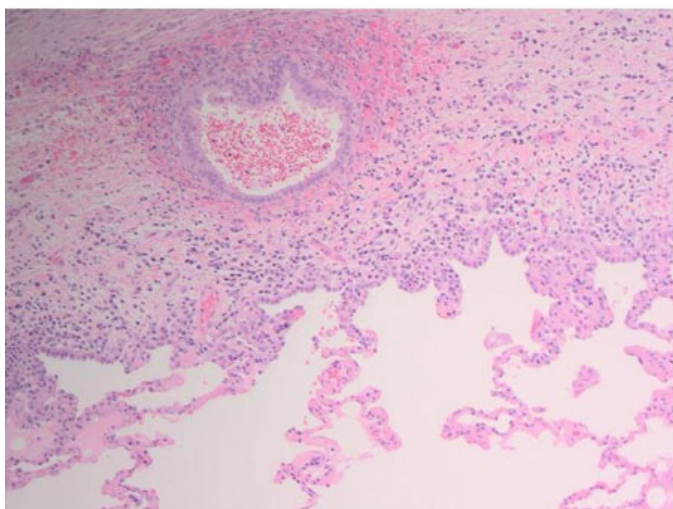


Figure 3: Micrograph at ×100 showing endometrial glands and stroma surrounded by hemorrhage and fibrosis. H&E stain.

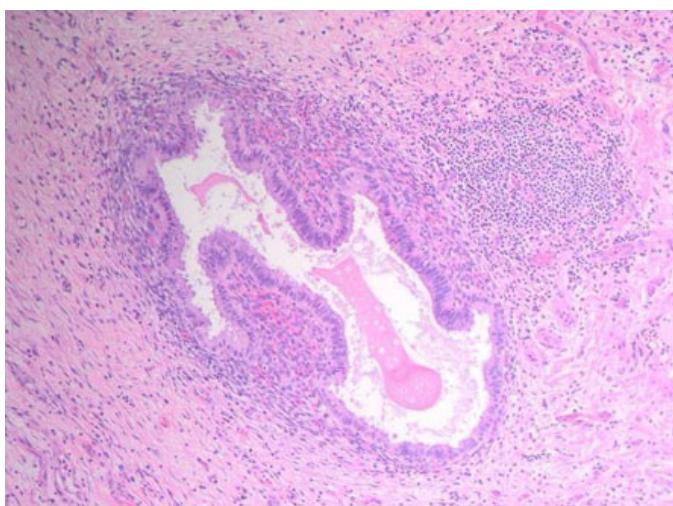


Figure 4: Micrograph at ×100 showing endometrial glands with stroma and hemorrhage. H&E stain.

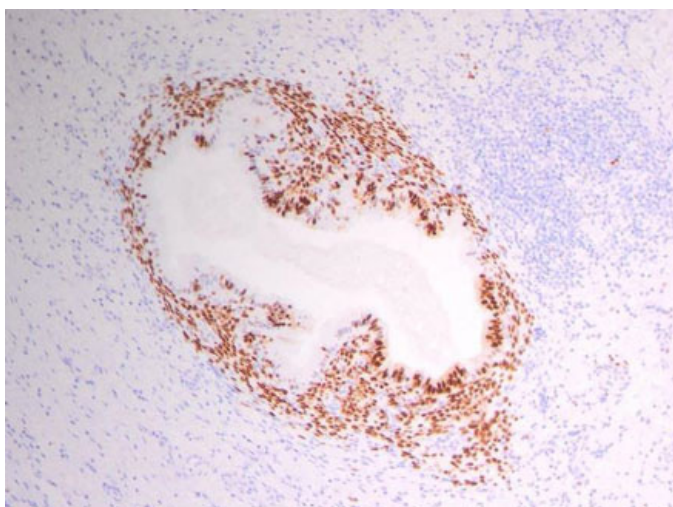


Figure 5: Micrograph at ×100 showing endometrial glands and stroma staining with estrogen receptor.

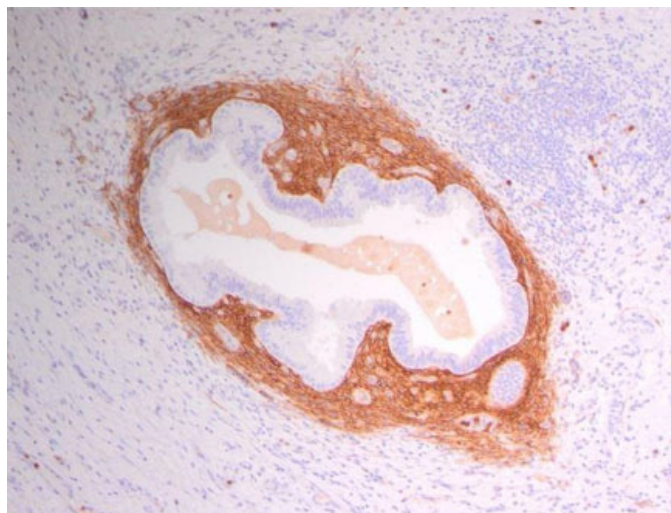


Figure 6: Micrograph at ×100 showing endometrial stroma staining with CD10.

DISCUSSION

Risk factors associated with catamenial pneumothorax include previous pelvic surgery, uterine scraping, infertility, mean age of 32–35 years, and symptoms of pelvic endometriosis [2]. Screening with these factors may enhance the diagnostic yield [3]. Our index case similarly was 34 years and had a history of infertility and had undergone 2 laparoscopic pelvic procedures prior to the presentation of catamenial pneumothorax. The typical clinical manifestation of catamenial pneumothorax involves spontaneous pneumothorax preceding or in synchrony with menses, usually presenting with pain, shortness of breath and cough. Scapular and or thoracic pain preceding or in synchrony with menses, history of previous episode(s) of spontaneous pneumothorax with or without previous surgical interventions is highly suspicious [4, 5]. Most of the cases of catamenial pneumothorax are right-sided, similar to this case but some can be left or bilateral. The pathogenesis of this disease entity is likely multifactorial. Several theories hypothesize the causation of this condition including the retrograde menstruation theory, coelomic metaplasia theory, lymphatic and hematogenous dissemination theory, and the prostaglandin theory [6].

A detailed history and physical examination are important prerequisites to the diagnostic work-up. The work-up is composed of imaging modalities such as chest X-ray (CXR), computed tomography (CT), and magnetic resonance imaging (MRI) [7]. Chest X-ray and CT chest are the most sensitive in detecting pneumothorax. Radiological findings include pneumomediastinum, pneumoperitoneum, ground-glass opacities, bronchial-wall thickening, thin-walled cavities within the lung parenchyma, or bullous formation [8, 9]. Chest X-ray in our case revealed a right-sided moderate to large hydropneumothorax and a chest CT revealed bilateral pleural nodularity in keeping with pleural-based

endometriosis deposits and right basal atelectatic changes. Interventional procedures like bronchoscopy, video laparoscopy, and video-assisted thoracic surgery are also included in the work-up regimen.

The gold standard of surgical intervention is video-assisted thoracic surgery (VATS) [10]. Depending on the variability of the case presentation, alternative procedures may also be performed including thoracotomy, and pleurodesis (chemically or surgically). Most commonly reported intraoperative findings included diaphragmatic lesions (38.8%), endometriosis of the visceral pleura (29.6%), discrete lesions such as bullae, blebs, or scarring (23.1%) and no findings (8.5%) [3]. In this case, there were adhesions seen between the right lung and lateral chest wall with some pleural abnormality; however, no overt diaphragmatic lesions or endometriosis of the lung pleura were observed intraoperatively.

Thoracic endometriosis has been considered as histologically “proven” after identification of endometrial stroma and glands in the thoracic lesions and as “probable” after identification of stroma only [4, 5, 11–13]. In our case, histologic examination showed endometrial glands and stroma surrounded by hemorrhage as well as fibrosis of the pleura; which was confirmed by immunohistochemical stains.

As far as treatment is concerned, VATS in conjunction with hormonal therapy for at least six months has been found to show an improvement in prognosis and a decrease in the possibility of a recurrence of catamenial pneumothorax [14]. Medical management includes hormonal therapy in the form of gonadotropin releasing hormones and aromatase inhibitors preferably for a period of 6–12 months. Similarly, our index case underwent VATS and was treated with Leuprolide for one-year post-surgery with no recurrence of symptoms of thoracic endometriosis.

CONCLUSION

There should be a high index of suspicion of catamenial pneumothorax in women of reproductive age who present with pneumothorax during or around the perimenstrual period. Adequate sampling of lung wedge resections and careful microscopic examination, followed by confirmation with immunohistochemistry play important roles in the diagnosis of this rare condition.

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Author Contributions

Oluwaseun Adelekan – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Moaz Alowami – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Pooja Vasudev – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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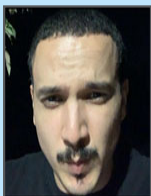
ABOUT THE AUTHORS

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Oluwaseun Adelekan is a resident doctor in the Department of Diagnostic and Clinical Pathology at McMaster University in Hamilton, Ontario, Canada. She earned the undergraduate degree (M.B.B.S) from the College of Health Sciences, University of Ilorin, Ilorin, Nigeria and postgraduate degree (FMCPATH) from the National Postgraduate Medical College of Nigeria. She has published five research papers in national and international academic journals. Her research interests include hematopathology, blood transfusion and gynecological pathology.

Email: Oluwaseun.adelekan@medportal.ca



Moaz Alowami is currently a research assistant at McMaster University as well as a clinical assistant at a walk-in clinic at Hamilton, Ontario, Canada. He earned his undergraduate degree (MBBS/MD) from Libyan International Medical University in Benghazi, Libya. He has published a total of 3 research papers so far in national and international academic journals. His research interests include pathology, internal medicine, and primary care medicine. He intends to pursue residency training in Canada.

Email: Drmoazalowami95@gmail.com



Pooja Vasudev is assistant professor at Department of Pathology and Molecular Medicine, McMaster University, Hamilton, Ontario, Canada. She earned the undergraduate degree (M.B.B.S.) from University of Delhi, Delhi, India and postgraduate degree (M.D. Pathology) from Department of Pathology/Punjab University, Chandigarh, India, followed by residency in Anatomical Pathology at McMaster University, Hamilton, Ontario, Canada. Currently, she is the Fellowship Director for the Breast Pathology Fellowship program, McMaster University. She has published numerous research papers in national and international academic journals. Her research interests include breast carcinoma special types, breast biomarkers, and unusual case reports related to skin and lung pathology. She intends to pursue more extensive research in breast biomarkers reporting in future.

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