CASE REPORT

Catamenial Pneumothorax: Breathing Through The Menstrual Cycle

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ABSTRACT

Catamenial pneumothorax (CP) is a unique disease related to thoracic endometriosis. It is often diagnosed in females of reproductive age presenting with recurrent spontaneous pneumothorax aligned with their menstrual cycle. We present a case of a woman presenting with recurrent episodes of dyspnoea coinciding with her menstrual cycle. Radiographic imaging confirmed a recurrent pneumothorax, and thoracoscopy with a pleural biopsy revealed diaphragmatic fenestrations and thoracic endometriosis. Multiple approaches, including medical pleurodesis, surgical pleurodesis, and hormonal therapy were used in her treatment. She recovered well after started on continuous oral Dienogest, and she has been asymptomatic for three years. This case illustrates the importance of recognizing the cyclical pattern of symptoms and a thorough diagnostic workup to confirm CP. This ensures timely management, which includes hormonal therapy and surgical intervention.

INTRODUCTION

Catamenial pneumothorax (CP) is a rare condition of recurrent spontaneous pneumothorax that predominantly affects women of childbearing age, occurring 48 to 72 hours before or after menstruation. About 3-6% of women presenting with spontaneous pneumothorax are believed to have an underlying catamenial cause, and 50-84% of them have concomitant pelvic endometriosis.[1] The incidence of CP may be underreported due to frequent misdiagnosis as spontaneous pneumothorax, mainly due to failure to associate the incidents with patients' menstrual cycle and not performing routine intraoperative diaphragm inspection.[2] A correct diagnosis of CP is vital for the patient, as hormonal therapy plays a role along with surgical management in preventing the recurrence.[3] In this report, we describe a case of CP, the diagnostic approach, multi-disciplinary team management involved, and the importance of a detailed history in a woman of reproductive age presenting with recurrent pneumothorax, for early detection of CP and to prevent its recurrence.

CASE PRESENTATION

A 41-year-old female initially presented to the Emergency Department (ED) during the early COVID-19 pandemic era with a complaint of shortness of breath and a non-productive cough for two days. She had no prior medical illness, and this was her first presentation for the complaint. There was no associated chest pain, palpitations, sore throat, fever or headache. Examination at the time showed that she was afebrile, but tachypneic and tachycardic with a respiratory rate of 34 breaths per minute and a pulse rate of 118 beats per minute. Her oxygen saturation was 94% under room air. Chest auscultations revealed reduced breath sounds on the right side, but no abnormal lung sounds were noted. She was started on a venturi mask with a fraction of inspired oxygen ($Fi0_2$) of 40%, and her oxygen saturation increased to 97%.

Chest radiograph revealed a right pneumothorax (Figure 1A); thus, a chest tube was inserted. Computed tomography (CT) of the thorax showed a small right

interlobar pneumothorax. Her COVID-19 swab test was negative. Due to her symptoms and in view of the current COVID-19 pandemic at the time, coupled with a negative COVID-19 swab test, she was treated as possible atypical pneumonia and started on oral Azithromycin and Oseltamivir. Her chest tube was removed after three days, and she was discharged home well after five days of admission. A follow-up respiratory clinic appointment was given in three months.

However, she returned to the ED with similar symptoms six weeks later. Chest radiograph at the time showed a right pneumothorax. A pigtail catheter was inserted since it has fewer complications and associated with shorter hospital stay (Figure 1C). The chest radiograph taken post -procedure shows an expanded lung field with a small residual ring pneumothorax in the apical region (Figure 1D). Therefore, medical pleurodesis with bleomycin was performed via the pigtail catheter on the fourth day of admission. A repeated CT scan of the thorax showed no new lesions. During this admission, further history revealed that both episodes of pneumothorax occurred within 72 hours of the onset of her menstrual cycle. Her menstrual cycle was usually regular every 28-30 days with the heaviest day occurring on day three of menses. She never has blood clots during her cycles and there was no associated dysmenorrhea or menorrhagia. She also denied any history of dyspareunia or pelvic pain during or around her menstrual cycle. Based on her history and presentation, a possibility of catamenial pneumothorax was very likely and she was referred to the gynaecology team for further management. However, since her pneumothorax had been stabilized and due to lack of personnel during the COVID-19 pandemic, she was only given an outpatient gynaecology clinic appointment.

Unfortunately, she experienced similar symptoms a week later and was readmitted. Since the previous medical pleurodesis had failed, she was then referred to the cardiothoracic team for further management. A thoracoscopy was performed, and it revealed multiple perforations at the centre of the diaphragm. Biopsies were taken from several affected sites on the diaphragm before surgical pleurodesis was carried out. Her symptoms improved after the procedure, and she recuperate well. However, on day five after the procedure, she experienced another episode of pneumothorax once the chest tube was removed. Thus, a mini thoracotomy was carried out to perform mechanical pleurodesis and plication of the central tendon of the diaphragm. She was started on continuous oral Dienogest 2mg and was well at discharge a few days later. The biopsy result of her diaphragm tissue showed endometrial stromal cells, suggesting endometriosis thus confirming the diagnosis of CP. She was advised to continue oral progestin until menopause. She claimed to have irregular menstrual cycle with occasional per vaginal spotting since started on continuous progestin. Otherwise, she is well and still under gynaecology clinic follow up and has not experienced any symptoms of pneumothorax or endometriosis since her last admission three years ago.

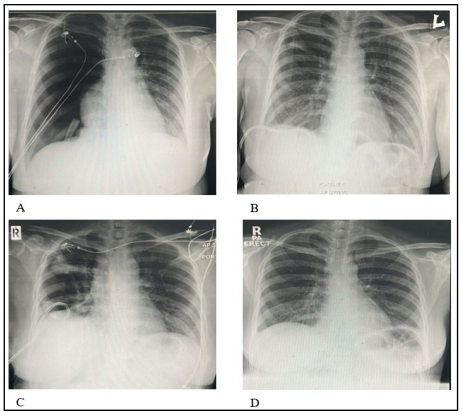

Figure 1: Image in 1A shows right pneumothorax (yellow arrow) and image 1B shows expanded lung field after chest tube drainage. Image 1C shows recurrent right pneumothorax (yellow arrow) after six weeks, while image 1D shows fully expanded lung post pig-tail catheter insertion.

DISCUSSION

CP is a rare cause of spontaneous pneumothorax which occurs in relation to the perimenstrual period. It accounts for 7.3% to 36.7% of all incidences of pneumothorax among women of reproductive age rendering it a clinically significant condition.[4] CP should be suspected in women in the reproductive age group who presented with recurrent spontaneous pneumothorax occurring within 72 hours before or after the onset of menstruation especially if they are known to have endometriosis.[5] CP is one of the entities in thoracic endometriosis syndrome (TES) which refers to the presence of endometrial tissue within the lung parenchyma or on the diaphragm and pleural surfaces causing catamenial haemothorax, haemoptysis, and pulmonary nodules.[6] Among these, CP

is the most frequent presentation and 60% of the cases are associated with pelvic endometriosis.[7] However, in our case, the patient had no known history of endometriosis and never had any prior symptoms nor sonographic evidence of pelvic endometriosis making the possibility of catamenial pneumothorax more remote initially.

The diagnosis of CP requires a comprehensive approach, starting with a detailed patient history to identify the cyclical nature of symptoms. Women with CP typically present with acute pleuritic chest pain and dyspnoea, symptoms that recur in association with their menstrual periods.[1, 6] The temporal relationship between symptom onset and menstruation is a key diagnostic clue that should prompt further investigation. Despite its distinctive cyclical pattern, CP often remains underdiagnosed due to a lack of awareness among healthcare providers and the rarity of the condition. As in this case, the first presentation was treated as spontaneous pneumothorax secondary to atypical Covid-19 related pneumonia as the association with her menstrual cycle was not made. She was also unfortunate since her first presentation occurred during Covid pandemic where the infection may be presented in many guises, hence, the suspicion of Covid related diagnosis. The establishment of temporal relationship between menstrual period and the occurrence of pneumothorax is the only clue for CP since there are no specific signs to distinguish it from other causes of pneumothorax.[8]

Radiographic imaging plays a limited role in the diagnosis and management of CP. The chest X-ray can confirm the presence of pneumothorax and evaluate the extent of lung collapse, but it may not identify small diaphragmatic defects or endometrial implantations.[4] On the other hand, CT scans and Magnetic Resonance Imaging (MRI) could show small diaphragmatic defects, called "air-filled bubbles", thus providing a better value in the diagnosis of CP.[8] A CT scan is usually performed first, as it is readily available. However, MRIs are more sensitive in detecting soft tissue lesions, especially diaphragmatic endometriosis.[4, 8] Both imaging modalities have a higher sensitivity rate if performed during menstruation.[8] Nevertheless, the imaging can only aid to confirm the diagnosis if the initial suspicion has been established. In this case, the role of imaging did not help much in establishing the diagnosis since the CT thorax only showed a right interlobar pneumothorax and not able to identify the diaphragmatic defect.

Once diagnosis has been established, the goal of the management of CP includes treating the acute illness and preventing the recurrence of the condition. Acute management of CP is similar to treating spontaneous pneumothorax. Long-term treatment options include a combination of medical, hormonal, and surgical therapy.[3] Pleurodesis of the lung is an option to prevent recurrence and can be achieved by both chemical and surgical methods.[1] Chemical pleurodesis is usually achieved by inserting a sclerosant through the chest tube aiming to prevent recurrent pneumothorax by causing chronic inflammation and scarring of the pleural surface.[1, 4]However, this method alone without concurrent surgical interventions results in a higher failure rate as illustrated by our patient who returned with a third episode of pneumothorax in just a week after a failed medical pleurodesis.

Video-assisted thoracic surgery (VATS) is the gold standard diagnostic tool for CP and may also function as a treatment modality.[8] It is a minimally invasive surgery that can be used to perform surgical pleurodesis, diaphragm repair, and bleb resections.[4] It has the advantage of smaller incisions, less post-operative pain, and fewer complications compared to thoracotomy which is the other surgical method widely used in the management of CP. Thoracotomy allows a wider surgical space and is useful in repairing diaphragmatic perforations and fenestrations in CP as demonstrated in our case after failed medical pleurodesis.[4] The repair is done by placing mesh over the diaphragm which promotes tissue in-growth thus providing scar tissue over the diaphragm.[4] Our patient underwent a mini-thoracotomy procedure during which her diaphragmatic fenestrations were repaired by mesh placement and enhanced with talc pleurodesis.

Hormonal therapy plays a crucial role in the treatment of CP. It reduces the risk of recurrence by suppressing

menstruation and the associated thoracic endometrial activity.[3] The maintenance of an amenorrhoeic state is vital in preventing the recurrence of CP. One study reported that resumption of menstruation leads to the recurrence of pneumothorax.[9] There are various classes of medications available for this purpose, including combined oral contraceptive pills, gonadotrophin-releasing hormone analogues (GnRHa), progestins, and aromatase inhibitors.[9]

Usually, GnRHa is used as first-line therapy as they are highly effective in suppressing the hypothalamic-pituitary-ovarian axis and the growth of endometrial cells. However, prolonged use of GnRHa may cause menopausal-like symptoms and loss of trabecular bone density. Thus, their use is limited to six to twelve months.[10] In view that our patient will be needing a long-term use of hormonal treatment, the GnRHa was not the best choice for her post operative treatment. On the other hand, progestins are found to be of similar efficacy as GnRHa but with a lower incidence of hypoestrogenic side effects.[9] Hence, continuous progestin was the hormone of choice in our patient who required effective long term treatment. Other issues which deserve consideration in choosing the type of hormonal treatment is the patient's fertility wishes. In our case, the patient has completed her family. Thus, being on continuous progestin is not an issue for her. However, in cases where patient wishes to conceive, the treatment might have to be changed to intermittent hormonal therapy to allow windows for fertility attempts.

The duration for hormonal treatment varies depending on the severity of endometriosis. Study found that 4mg of progestin daily was successful in maintaining amenorrhoea and preventing recurrent CP at a six-month follow-up.[9] Most studies suggest a duration of six to twelve months, but patients with recurrence of CP like our patients may require a longer duration. Patients with CP especially those requiring long-term hormonal treatment will need to undergo surveillance at least biannually for clinical symptoms of recurrence and to monitor for side effects of the medication.

CONCLUSION

CP is a complex and rare condition that demands a comprehensive and multidisciplinary approach for effective diagnosis and management. This case report highlights the significance of recognizing CP in women of reproductive age who present with recurrent pneumothorax associated with their menstrual cycle.

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CONFLICT OF INTEREST

The authors have no potential conflict of interest relevant to this case report to declare.

INSTITUTIONAL REVIEW BOARD (ETHIC COMMITTEE)

The authors certify that all required patient consent forms have been received.

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AUTHOR'S CONTRIBUTION

SL wrote the original draft and obtained the images. NSI conceptualized the case report, its aims and reviewed the initial draft. RAR revised it critically for intellectual content and edited the case for final submission. All the authors critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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