Catamenial pneumothorax: A rare cause of recurrent pneumothorax

Waqas Jehangir, Jay Harman, Nneka Iroka, Abdalla Yousif

ABSTRACT

Introduction: Primary spontaneous pneumothorax is a common clinical occurrence. Although primary spontaneous pneumothorax is twice as common in men as in women. The recurrence rate is significantly higher in women. The two primary causes for recurrence in women are catamenial pneumothorax and endometriosis related pneumothorax. In the past, catamenial and/or endometriosis related pneumothorax were greatly underdiagnosed. The incidence has increased in the past decade because it is more easily recognized today. Spontaneous pneumothorax is a lung compression that occurs spontaneously due to air in the pleural space in a patient with no underlying lung disease. It can occur in men or women but occurs most often in men. It occurs through many different causes. Catamenial pneumothorax is a spontaneous pneumothorax that occurs at the time of menses in a woman that allows air to enter the thoracic space. Non-catamenial endometriosis related pneumothorax is a spontaneous pneumothorax that occurs when endometrial tissue ascends through diaphragmatic defects to the pleural space and allows air to enter. It can occur at any time and not just during the menses in a woman. Catamenial pneumothorax and noncatamenial endometriosis related pneumothorax are independent entities and are not synonyms. They may occur simultaneously but do not necessarily have to occur at the same time.

Case Report: We present a case of recurrent pneumothorax diagnosed as catamenial pneumothorax in an otherwise healthy 34-year-old female.

Conclusion: The percentages of catamenial pneumothorax are still unclear but it should be suspected and affectedly treated.

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Keywords: Primary spontaneous pneumothorax, Catamenial pneumothorax, Endometriosis, Video assisted thoracoscopic surgery

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INTRODUCTION

Primary spontaneous pneumothorax was first described in 1932 as a separate medical condition that occurs in patients without lung disease [1]. The incidence is thought to be increasing and the recurrence rate is between 20–60% [1]. The male to female ratio in primary spontaneous pneumothorax is 2:1 while the recurrence rate is significantly higher in women [1]. The term recurrent does not define the exact number of episodes although the mean is very high: five episodes with as many as 10 episodes in some cases [2]. At least two episodes are...
required to be considered recurrent pneumothorax [3]. Therefore, the occurrence of recurrent pneumothorax is a significant problem especially in women. Catamenial pneumothorax and/or endometrial related pneumothorax are one of the main causes of recurrent pneumothorax in women. Although known since the 1950s, catamenial pneumothorax was considered an extremely rare condition and was greatly underdiagnosed [2]. Although catamenial and/or endometriosis related pneumothorax are recognized more now than in the past, their real frequency remains unclear [4]. The signs and symptoms of recurrent pneumothorax are the same as for other kinds of pneumothorax—chest pain, shortness of breath, and cough [2].

CASE REPORT

A 34-year-old Hispanic female non-smoker with no significant past medical history stated that she had been having pain in the scapula four days ago which was associated with shortness of breath. She described the pain as a sharp, constant, radiating to the front and back of the right side of the chest. She also had shortness of breath which was associated with chest pain and dry cough. She was diagnosed with pneumonia by her primary medical doctor and she was prescribed antibiotics and pain medications which did not help her. She gave a history of spontaneous pneumothorax on her right side six months ago. She denied any fever, chills, nausea, and vomiting but stated that the pain gotten progressively worse. She did not have a history of tuberculosis or endometriosis. She had breast lifting seven years ago. Her last menstrual period was three days ago. She had three abortions which were induced and she has two babies. On physical examination, she was in mild respiratory distress and vital signs were temperature 98°F, blood pressure 127/82 mmHg, pulse 84/min, respiration rate 20/min, and PO2 96% on 2 litre/minute of oxygen per nasal canula. Lung examination revealed decreased air entry on the right side and decreased breath sounds on the right side. Rest of the physical examination was unremarkable. Laboratory data showed white blood cell count 10.4x10^3/μL, hemoglobin 13 g/dL, hematocrit 41%, platelets 343x10^3 K/uL, neutrophils 68%, lymphs 21%, BUN 8 mg/dL, creatinine 0.7 mg/dL, albumin 3.9 mmol/L, potassium 3.9 mmol/L, chloride 99 mmol/L, CO₂ 26 mmol/L. Chest X-ray showed 90% of pneumothorax (Figure 1). She was admitted and emergency right closed thoracostomy drainage done and mechanical pleurodesis was performed. Alpha 1 antitrypsin later came to be 106.00 IU/mL (90–200 IU/mL). Patient was diagnosed with catamenial pneumothorax. She was discharged home and has remained free of recurrence six months after re-treatment.

DISCUSSION

While men are twice as likely to have primary spontaneous pneumothorax as women, whereas women are much more likely to have a recurrence. The current theory is that a woman’s menstrual cycle and/or endometriosis play a role in this reoccurrence. The mean age for recurrent pneumothorax in a woman is 32 years [5].

Catamenial pneumothorax is a recurrent pneumothorax occurring 24 hours before and up to 72 hours after the onset of menses [4]. There are two hypotheses concerning the causes of catamenial pneumothorax. The first hypothesis is that the open connection between the atmosphere and the peritoneal cavity allows air to enter the thoracic cavity through diaphragmatic fenestrations and porosities [5]. In the menstrual period the cervical mucus plug is absent, thus permitting communication between the peritoneal cavity and the outside through the uterus and the fallopian tubes. Air may be forced to enter the peritoneum by uterine contractions, physical exercise, or sexual intercourse. The air then reaches the pleural space through diaphragmatic defects because of negative intrathoracic pressure [2]. One argument supporting the theory of transdiaphragmatic passage of air is the observation that recurrent catamenial pneumothorax may be prevented by tubal ligation. The second hypothesis is that prostaglandin P₂, a potent constrictor of bronchioles may destroy alveolar tissue causing alveolar rupture and...
pneumothorax [5]. In the menstrual period, many women have increased levels of prostaglandin F2-a. Catamenial pneumothorax is unilateral and right sided in almost all instances [2].

The other hypotheses are that endometriosis plays a role in the reoccurrence of pneumothorax. The endometrial tissue reaches the thoracic cavity through auto transplantation to ectopic sites through lymphatic or vascular embolization or after retrograde menstruation [4]. The endometrial tissue can reach the thoracic cavity through diaphragmatic defects possibly caused by endometriosis [5]. This mechanism explains why there is right sided predominance in recurrent pneumothorax. The peritoneal fluids along with air and endometrial tissue exit from the pelvis along the right paracolic gutter up to the right subphrenic space [2, 3] and then through the diaphragmatic fenestrations and porosities. Endometriosis related pneumothorax is considered proven when endometrial glands and stroma are demonstrated by immunohistochemistry staining [4]. Thus endometriosis related pneumothorax can occur in the intermenstrual period [2] as well as the menstrual period, while catamenial pneumothorax only occurs in the menstrual period 24 hours before and up to 72 hours after menses.

The diagnosis of thoracic endometriosis has improved over the past two decades because of Video Assisted Thoracoscopic Surgery (VATS). The VATS is considered the gold standard for both definitive diagnosis and surgical treatment of catamenial and/or endometriosis related pneumothorax [5] and has been applied since 2000 [3]. Some clinicians influenced by the 50% recurrence rate of catamenial and/or endometriosis related pneumothorax advocate an aggressive approach with early surgical treatment [1]. Diaphragmatic involvement by either endometrial tissue or perforations is probably best treated by diaphragmatic resection. Talc pleurodesis is recommended instead of pleural abrasion because of higher recurrence rate with pleural abrasion [2]. Current studies have concluded that surgery has better results than hormone treatment in preventing recurrence. The best results have been obtained using surgery followed by either GnRH agonists or the antigonadotropic progestins cyproterone acetate for six months to induce amenorrhea [2]. If the stapling of the diaphragmatic lesions, the pleurodesis, or the hormone treatment does not prevent recurrence, then hysterectomy and bilateral salpingo-oophorectomy are the treatments of last resort [6].

In our case, the patient had all the classical symptoms of catamenial pneumothorax. She presented with SOB along with chest pain and a cough. Her last menstrual period was three days previously at the time she first had the pain. The lung exam and X-ray revealed a right sided pneumothorax. She had a history of spontaneous pneumothorax six months prior to this. For treatment the patient had a thoracostomy drainage done and mechanical pleurodesis. She has remained free of recurrent pneumothorax at six months.

CONCLUSION

The percentages of catamenial and/or endometriosis related recurrent pneumothorax are still unclear so additional research is needed to clarify this. But it should be suspected and affectedly treated. Operations for catamenial and/or endometrial related pneumothorax have practically zero mortality and no significant morbidity.

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Author Contributions

Waqas Jehangir – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Jay Harman – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Nneka Iroka – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Abdalla Yousif – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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