



## Recurrent Catamenial Pneumothorax after Failed Doxycycline Pleurodesis

Anthony McCloud<sup>1</sup> and Osita Onugha<sup>2\*</sup>

<sup>1</sup>Department of Thoracic Surgery, Western University of Health Sciences, College of Osteopathic Medicine of the Pacific Pomona, USA

<sup>2</sup>Department of Thoracic Surgery Providence St. John's Health Center, John Wayne Cancer Institute, Santa Monica, USA

### Abstract

Catamenial Pneumothorax (CP) is a rare type of Spontaneous Pneumothorax (SP) that affects women, especially those of reproductive age. CP is characterized by thoracic endometrial lesions and diaphragmatic fenestrations leading to pneumothorax and is often in close relationship to a woman's menses. We report a female of reproductive age that presents with recurrent.

### Introduction

Spontaneous pneumothorax is classified as either Primary Spontaneous Pneumothorax (PSP) or Secondary Spontaneous Pneumothorax (SSP) that are defined by the underlying lung condition. Patients with PSP lack underlying lung conditions but often have findings of ruptured blebs or bulla resulting in pneumothorax. Patients with SSP present with lung conditions or disease that resulting in pneumothorax. Endometriosis is characterized by the growth of endometrial glands and stroma outside of the uterine cavity [1]. Endometriosis is typically named by the cavity in which the exogenous endometrial tissue is found; for example, pelvic endometriosis if found in the pelvis and thoracic endometriosis if found in the thoracic cavity [1]. With thoracic endometriosis there are four clinical presentations to be aware of: catamenial hemothorax, catamenial hemoptysis, endometriotic pulmonary nodules, and catamenial pneumothorax; these groups of conditions constitute Thoracic Endometriosis Syndrome (TES). While it is unclear the frequency of TES, CP is considered the most common form of TES as evidenced by the Joseph and Sahn series of 110 TES cases with 73% being CP [2].

### OPEN ACCESS

#### \*Correspondence:

Osita Onugha, Department of Thoracic Surgery, Providence St. John's Health Center, John Wayne Cancer Institute, 2121 Santa Monica Blvd, Cardiothoracic Outpatient Clinic, Santa Monica, CA 90404, USA, Tel: (310) 829-8618;

E-mail: [osita.onugha@providence.org](mailto:osita.onugha@providence.org)

Received Date: 23 Jul 2019

Accepted Date: 30 Jul 2019

Published Date: 02 Aug 2019

#### Citation:

McCloud A, Onugha O. Recurrent Catamenial Pneumothorax after Failed Doxycycline Pleurodesis. *Clin Surg.* 2019; 4: 2531.

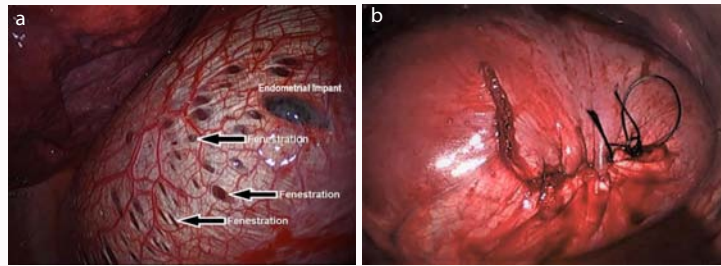
Copyright © 2019 Osita Onugha. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

### Case Presentation

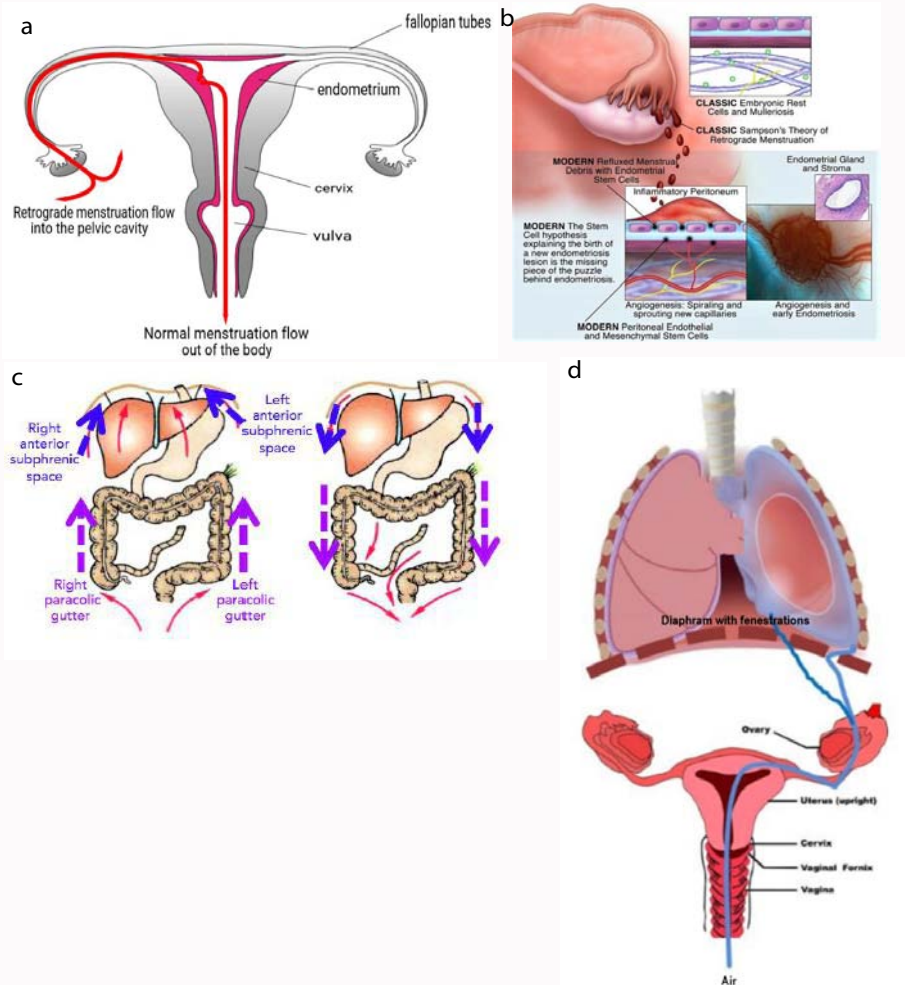
We report a woman in her late thirties with a history of Gastroesophageal Reflux Disease (GERD), fibroids, endometriosis, and recurrent Spontaneous Pneumothorax (SP). She presented to the ER six days after the onset of her menses with complaints of Shortness of Breath (SOB) and chest pain upon lying flat. A CXR was performed and she was found to have a right pneumothorax. She was taken to the OR for bronchoscopy; right VATS wedge resection, and doxycycline pleurodesis. On intra-operative inspection of the diaphragm, and endometrial implant was seen with several (more than 45) diaphragmatic fenestrations (Figure 1a). The endometrial implant was biopsied and confirmed to be endometrial tissue, consistent with Catamenial Pneumothorax (CP). The diaphragmatic fenestrations were closed using 0-ethibond suture (Figure 1b). Finally, pleurodesis with 500 mg of doxycycline was performed and the chest was drained via chest tubes. Upon completion of the operation the patient was in stable condition. The patient was discharged home on POD 2 with no evidence of pneumothorax on follow up. Two months after discharge, she underwent a laparoscopic hysterectomy for uncontrolled pelvic endometriosis resulting in a post-operative pneumothorax. A chest tube was placed and Chest X-Ray (CXR) was performed and confirmed a persistent right pneumothorax despite chest tube placement. She was taken back to the operating room for a right VATS talc pleurodesis due to the previously failed doxycycline pleurodesis. Upon inspection of the thoracic cavity adhesions were seen between the lung and the diaphragm, however, no adhesions were seen between the lung and the chest wall. As a result, four grams of talc was used for pleurodesis. The chest was drained via chest tubes and the patient was in stable condition at the end of the procedure. She has had no recurrence in symptoms.

### Discussion

In this paper we present a reproductive age female with recurrent pneumothorax. Though it was



**Figure 1a:** Pre-repair: Endometrial implant present in the diaphragm confirmed by presence of endometrial glands and stroma. Additionally, multiple fenestrations are visible in the diaphragm. Post-repair: Staple line from resection of endometrial implant and suture closure of diaphragm fenestrations.



**Figure 2a:** Diagram of retrograde menstruation as proposed by Sampson's Theory. **2b:** Pathophysiology of endometriosis as a result of retrograde menstruation. Additionally, angiogenesis is depicted at the site of the endometrial implant which is a proposed mechanism for travel of endometrial tissue outside of the pelvic and peritoneal cavity. **1c:** Depiction of fluid flow (red arrows) through the peritoneal cavity demonstrating how endometrial tissue may travel to the diaphragm via these gutters. **2d:** Demonstrates how air can enter the pleural space through diaphragm fenestrations via the female reproductive system. This results in the formation of a CP.

not initially believed her pneumothorax had any relationship to her menses, intraoperative findings suggested CP. This is consistent with the high temporal variability between onset of menses and clinical manifestation of CP [3]. As such, CP is typically not diagnosed until surgery. This may explain why studies show an increasing incidence of CP in women with pneumothorax. It has therefore been accepted that the best way to confirm a diagnosis of CP is to have the presence of endometrial tissue with glands and stroma present in the thorax [1]. Analysis of the implant in our patient's diaphragm proved to be

endometrial tissue allowing for confirmation of the diagnosis of CP despite the onset of her symptoms occurring six days after the start of her menses. This demonstrates and gives further evidence that the temporal relationship between the menses and CP is highly variable. The process by which endometrial tissue enters the thoracic cavity is still unclear, however there are three theories behind thoracic endometrial implants: lymphatic or venous transport, coelomic metaplasia, aberrant stem cells, and retrograde menstruation [4,5]. Of these, retrograde menstruation has risen as the dominant mechanism

for TES as described by Sampson's Theory of retrograde menstruation [4,5]. Retrograde menstruation is the process by which some of the menstrual fluids move opposite the normal direction of flow (Figure 2a). Instead of flowing towards the vaginal canal, menstrual fluids move further into the uterine cavity and into the fallopian tubes from which menstrual fluids can then enter the peritoneal cavity. Though retrograde menstruation has been shown to occur in most healthy women [6] the mechanism by which some women develop endometriosis is still unclear. Upon retrograde menstruation, endometrial tissue enters the peritoneal fluid which predominantly flows up the right para-colic gutter to the right sub-diaphragmatic area where the endometrial tissue implants into the diaphragm [7] (Figure 2b, 2c). This is the most common proposed mechanism of flow of endometrial tissue from the peritoneal cavity to the thoracic cavity.

This also explains why right CP is more common. Bleeding of the endometrial tissue during menses creates diaphragmatic defects known as fenestrations [4]. Without repair, these fenestrations allow further communication of endometrial tissue into the thoracic cavity. Furthermore, these fenestrations allow passage of air from the peritoneal cavity which has been proposed as a mechanism for CP [4] (Figure 2d). Other proposed mechanisms of CP include hormone induced contraction of bronchioles and blood vessels leading to rupture of blebs and alveoli leading to air flow into the pleural space. Lastly, and the most widely accepted, is the cyclical sloughing of endometrial implants in terminal bronchioles leading to air leak into the pleural space [4,7]. CP is commonly treated surgically by Video Assisted Thoracoscopy (VATS) [4]. Surgery primarily focuses on lung resection to remove blebs (Figure 2d), inspection and repair of diaphragm fenestrations, removal of endometrial implants, and pleurodesis [8]. Pleurodesis is achieved by pleural abrasion or chemical agents; commonly doxycycline or talc. However, studies have shown a lower recurrence of pneumothorax with talc pleurodesis over doxycycline pleurodesis in patients with CP [2,4,8]. Hormonal treatments such as oral contraceptive, androgen treatment, and Gonadotropin Releasing Hormone treatments (GnRH) have also been used as treatment to prevent recurrent CP but prove to be less effective and have shown to have higher rates of recurrence [2]. In this case initial pleurodesis was performed using doxycycline insufflation which was determined to have failed leading to another SP. This is consistent with findings of increased SP recurrence with doxycycline pleurodesis over talc pleurodesis [2,4,8].

Though it has not yet been agreed upon which chemical pleurodesis technique is most effective, we suggest that talc is used as the primary pleurodesis agent in patients with CP to prevent recurrent pneumothorax.

## Conclusion

The findings in this case further show the timing variability of CP in coordination to the menses. As such, clinicians should consider CP in women with SP and thoroughly inspect the diaphragm in all young women with a spontaneous pneumothorax. Furthermore, this case shows the potential inadequacies of doxycycline pleurodesis suggesting the use of talc pleurodesis as the preferred agent for pleurodesis in CP.

## References

1. Alifano M, Jablonski C, Kadiri H, Falcoz P, Gompel A, Camilleri-Broet S, et al. Catamenial and noncatamenial, endometriosis-related or nonendometriosis-related pneumothorax referred for surgery. *Am J Respir Crit Care Med.* 2007;176(10):1048-53.
2. Joseph J, Sahn SA. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. *Am J Med.* 1996;100(2):164-70.
3. Joseph-Vempilly, J. Clinical features, diagnostic approach, and treatment of adults with thoracic endometriosis.
4. Alifano M, Trisolini R, Cancellieri A, Regnard JF. Thoracic endometriosis: current knowledge. *Ann Thorac Surg.* 2006;81(2):761-9.
5. Schenken RS. *Endometriosis: Pathogenesis, clinical features, and diagnosis.* UpToDate, Waltham, MA: Walters Kluwer Health. 2016.
6. Halme J, Hammond MG, Hulka JF, Raj SG, Talbert LM. Retrograde menstruation in healthy women and in patients with endometriosis. *Obstet Gynecol.* 1984;64(2):151-4.
7. Shrestha B, Shrestha S, Peters P, Ura M, Windsor M, Naidoo R. Catamenial Pneumothorax, a commonly misdiagnosed thoracic condition: Multicentre experience and audit of a small case series with review of the literature. *Heart Lung Circ.* 2019;28(6):850-857.
8. Fournel L, Bobbio A, Robin E, Canny-Hamelin E, Alifano M, Regnard JF. Clinical presentation and treatment of catamenial pneumothorax and endometriosis-related pneumothorax. *Expert Rev Respir Med.* 2018;12(12):1031-6.