The ESRF Research Journal for Undergraduate Medical Students

Case Report

Volume 2 Issue 1

2024

Submission:
9th October, 2024
Acceptance:
13th October, 2024
DOI:10.5281/zenodo.13924562

Corresponding Author

Dr. Anupam Patra
Flat -1C, Sunny dew-2, Garia, Kolkata-700084.
e-mail:
dranupam.patra@gmail.com

Indexed in:
Google Scholar
BASE
Bielefeld Academic Search Engine

Citation: Roy S, Patra A. Pleural endometriosis: A rare cause of spontaneous haemopneumothorax. RJUMS 2024; 2(1):i-iii.

Available from: https://esrfrjums.co.in/index.php/main/article/view/48

Published by: Eureka Scientech Research Foundation, Kolkata.

Online access: https://esrfrjums.co.in

Pleural endometriosis:
A rare cause of spontaneous haemopneumothorax

Suchisubhra Roy,[1]
Anupam Patra,[2]

[1]*MBBS Student (2020 – 2025), Nil Ratan Sircar Medical College, Kolkata*

[2]*Post Doctoral trainee, Department of Pulmonary Medicine, Institute of Post Graduate Medical Education & Research, Kolkata*

Abstract

The presence of endometrial tissue in airways, pleura and lung parenchyma is called thoracic endometriosis syndrome (TES). It is a rare pathology, and typically consists of catamenial pneumothorax, haemothorax, haemoptysis, and pulmonary nodules. We report a case of a 37-year-old woman with thoracic endometriosis causing catamenial hemopneumothorax. The diagnosis was made on proper history, imaging, thoracoscopic biopsy and immunohistochemistry. The diagnosis of thoracic endometriosis is complicated and often delayed. TES should be suspected in a reproductive age woman with exacerbating symptoms during the menstruation.

Keywords: thoracic endometriosis, pleura, hemopneumothorax

INTRODUCTION

Endometriosis is defined as the presence of normal endometrial tissue outside the uterine cavity.[1,2] It is diagnosed in women of reproductive age. Endometriosis most commonly affects pelvic organs, but it can be found in extra-pelvic organs and tissues like airways, pleura and lung parenchyma.[3,4] The presence of endometrial tissue in or around the lung is called thoracic endometriosis syndrome (TES). Typically, it consists of catamenial pneumothorax, haemothorax, haemoptysis, and pulmonary nodules.[2,5] The first case of pleural endometriosis was described by Schwarz in 1938.[6] The first case of catamenial pneumothorax was described in 1958 by Mauer et al,[7] and the term "*catamenial pneumothorax*" was first introduced into the literature in 1972 by Lillington et al.[8]

CASE PRESENTATION

A 37 year old woman presented to our OPD with right sided chest pain for 3 months, dry cough for 3 months and dyspnoea for 2 months. She had a history of admission for same type of illness 1 year back. At that time right sided spontaneous pneumothorax was diagnosed on routine chest X-ray and was treated with intercostal tube drainage. There was no history of trauma, fever, haemoptysis, weight loss. Patient gave history of cyclical pain in right side of thorax during menstruation.

This time, on examination right sided pneumothorax was present, her SpO_2 was 91% on room air and respiration rate was 22 per minute. Laboratory tests and ECG did not show any abnormalities. Sequential chest x-ray revealed appearance and disappearance of pneumothorax. Blood laboratory findings revealed anaemia: haemoglobin-8.6 gm/dl, red blood cells-3.63 × 10^{12}/l. Chest x-ray showed hydropneumothorax on the right side (Fig. 1). 500 ml of haemorrhagic fluid from pleural cavity was drained and dyspnoea was relieved to some extent. CECT thorax revealed right sided pleural effusion. USG guided pleural fluid study was suggestive of haemothorax. Ultrasound of abdomen showed pelvic endometriosis. Fibreoptic bronchoscopy was non-contributory.

We performed rigid thoracoscopy and it revealed multiple violaceous patches seen along with multi-

-ple clusters of reddish spots (Fig.2). Histopathological examination confirmed the diagnosis of endometriosis. (Fig 3) Immunohistochemistry report came out to be CD 10 and ER, PR positive. The patient underwent gynaecological examination. A transvaginal ultrasound examination revealed pelvic endometriosis and was treated with gonadotropin-releasing hormone (GnRH) agonists. At present, the patient is under observation of chest physician and gynaecologist. The patient has no respiratory tract symptoms as well any abnormalities on imaging after 3 months follow up chest X-ray.

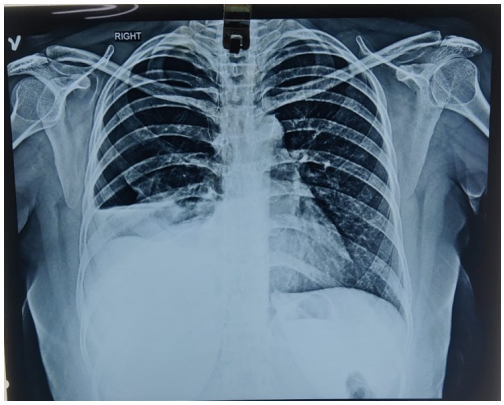

Fig 1: Chest X ray showing haemopneumothorax

Fig 2: Thoracoscopy showing violaceous patches with red spots

DISCUSSION

Endometriosis most commonly affects pelvic organs, but it can be found in extra-pelvic organs and tissues.[3,4] The presence of endometrial implants in airways, pleura and lung parenchyma is called thoracic endometriosis syndrome (TES).[5] It consists of catamenial pneumothorax, catamenial haemothorax, catamenial haemoptysis, and pulmonary nodules.[2,5] Most patients commonly have catamenial pneumothorax (73%), while catamenial haemothorax is present just in 14% of the cases, followed by 7% of haemoptysis and 6% of lung nodules.[9] In our case, the patient suffered from the right side hemopneumothorax. There are several different proposed theories for the formation of TE.[4,10] The first hypothesis is lymphatic or

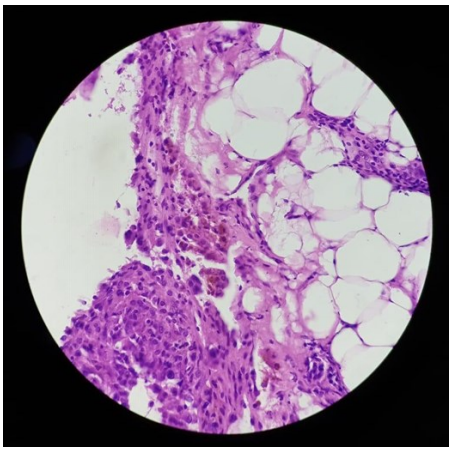

Fig 3: Histopathology findings on pleural biopsy

haematogenous embolization from the uterus,[4,10] the second presents coelomic metaplasia theory and the third proposes retrograde menstruation during which endometrial tissue from the uterus and fallopian tubes migrates via abdomen and congenital or acquired diaphragmatic defects into the pleural cavity. [10,11].

Caesarean section increases the risk of endometriosis. Therefore, constantly growing number of caesarean sections will be the main factor of increasing incidence of pelvic endometriosis as well as distant endometriosis, e.g. pleural and chest. Population studies by Haga et al., enabled development of point system risk assessment of pleural endometriosis. Predictive factors include: right sided pneumothorax, pelvic endometriosis, age above 31 years, non-smoking (sensitivity 93.5% and specificity 89.4% with the result of 12 points or more).[11] Pleural endometriosis is accompanied by chest discomfort and pain, which sometimes may by neglected by patients. [12]

Our patient confirmed the early onset of respiratory tract symptoms. Numerous hospital admissions to pulmonology wards emphasize repetitiveness and cyclic nature. Fallopian tube ligation is believed to be preventive of further dissemination of endometrial tissue via fallopian tube, and then through the fenestrations in the diaphragm into the chest.[10] Patient's induction into artificial menopause was correlated with regression of respiratory tract symptoms and arrest of pneumothorax recurrence. One should emphasize that restoration of natural menstrual cycle resulted in recurrence of dyspnoea and pneumothorax. It was caused by activation of ectopic endometrium within the lungs.

As in a case presented by Zeena Makhija et al., video-thoracoscopy (gold standard in diagnosis and treatment of pleural endometriosis) was performed with lobectomy and pleuradhesis (creation of adhesions between visceral and parietal pleura).[13] VATS procedure included close inspection of chest cavity, including diaphragmatic aspect, which did not show any abnormalities. Presence of visible fenestrations in diaphragm may explain the pathway of endometrial tissue is it was in case described above.[13] However, invisible micro-fenestrations may also be seen sites for the ectopic tissue.[6] The hypothesis about hemato and/ or lymphogenic dissemination is also

possible considering the fact of surgical intervention within the uterus (caesarean section) in the past.[8] The ultimate diagnosis and confirmation is based on the histopathological examination. H&E staining usually does not allow detecting foci of endometriosis. The recommended method to detect ectopic endometrial tissue is immunohistochemistry (CD 10, oestrogen and progesterone receptors).[14]

A characteristic finding was presence of eosinophilic inflammatory infiltrate and numerous macrophages. In a case of pleural endometriosis reported by Mesbah Rahman, routine H&E stain did not reveal endometrial structures but additional immunohistochemical examinations detected endometrial cells.[15] TES is a complex disease causing numerous diagnostic problems among clinicians as well as among pathologists. Spontaneous recurrent pneumothorax that accompanies menstruation in women with diagnosed pelvic endometriosis.[2,14] Diagnosis of TES requires proper history taking, histopathological examination and immunohistochemistry. The treatment options for TES are medical, surgical, and combined therapy. The target of medical treatment is focused on the suppression of ovarian oestrogen secretion. For this purpose, danazol and gonadotropin-releasing hormone (GnRH) agonists are used.[2,3,10,16] The diagnosis and appropriate therapeutic approach are especially important for successful treatment.

CONCLUSION

The diagnosis of thoracic endometrial syndrome (TES) is complicated and often delayed. TES should be suspected in a reproductive age woman with exacerbating symptoms during the menstruation. A great attention should be paid to the importance of taking a thorough history (especially careful gynaecological history), a comprehensive physical and radiological and histopathological examination.

CONFLICT OF INTEREST

Not Declared

FUNDING

Not Declared

REFERENCES

1. Machairiotis N, Stylianaki A, Dryllis G, Zarogoulidis P, Kouroutou P, Tsiamis N, Katsikogiannis N, Sarika E, Courcoutsakis N, Tsiouda T, Gschwendtner A. Extrapelvic endometriosis: a rare entity or an under diagnosed condition?. Diagnostic pathology. 2013 Dec;8:1-2.
2. Azizad-Pinto P, Clarke D. Thoracic endometriosis syndrome: case report and review of the literature. The Permanente Journal. 2014;18(3):61.
3. Hwang SM, Lee CW, Lee BS, Park JH. Clinical features of thoracic endometriosis: A single center analysis. Obstetrics & Gynecology Science. 2015 May 19;58(3):223-31.
4. Alwadhi S, Kohli S, Chaudhary B, Gehlot K. Thoracic endometriosis-a rare cause of haemoptysis. Journal of Clinical and Diagnostic Research: JCDR. 2016 Apr;10(4): TD01–TD02.
5. Suwatanapongched T, Boonsarngsuk V, Amornputtisathaporn N, Leelachaikul P. Thoracic endometriosis with catamenial haemoptysis and pneumothorax: computed tomography findings and long-term follow-up after danazol treatment. Singapore medical journal. 2015 Jul;56(7):e120–e123.
6. Schwarz O. Endometriosis of the lung. Am J Obstet Gynecol. 1938; 36: 887-889.
7. Maurer ER, Schaal JA, Mendez FL. Chronic recurring spontaneous pneumothorax due to endometriosis of the diaphragm. J Am Med Assoc. 1958; 168: 2013-2014.
8. Lillington GA, Mitchell SP, Wood GA. Catamenial pneumothorax. JAMA. 1972; 219: 1328-1332.
9. Joseph J, Sahn SA. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. The American journal of medicine. 1996 Feb 1;100(2):164-70.
10. Nwiloh J. Diaphragmatic patch: a useful adjunct in surgical treatment of recurrent catamenial hemothorax. Revista Portuguesa de Pneumologia (English Edition). 2011 Nov 1;17(6):278-80.
11. Joseph J, Sahn SA. Thoracic endometriosis syndrome: new observations from an analysis of 110 cases. Am J Med. 1996; 100: 164-170.
12. Ichiki Y, Nagashima A, Yasuda M. Surgical treatment of catamenial pneumothorax: Report of three cases. Asian J Surg. 2013; 38: 180-185.
13. Makhija Z, Marrinan M. A case of catamenial pneumothorax with diaphragmatic fenestrations. J Emerg Med. 2012; 43: e1-3.
14. Flieder DB, Moran CA, Travis WD, Koss MN, Mark EJ. Pleuropulmonary endometriosis and pulmonary ectopic deciduosis: a clinicopathologic and immunohistochemical study of 10 cases with emphasis on diagnostic pitfalls. Hum Pathol. 1998; 29: 1495-1503
15. Rahman M, Szafranek A, Ahmed Y. Catamenial pneumothorax: lessons learned and literature review. 2009; 6: 73-76.
16. S. Sevinc, S. Ünsal, T. Öztürk, et al., Thoracic endometriosis syndrome with bloody pleural effusion in a 28 year old woman, J. Pak. Med. Assoc. 63 (1) (2013) 114–116