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Case Report

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Air of Despair: Unveiling the Enigma of Dual Lung Collapse

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Introduction

Thoracic Endometriosis Syndrome (TES) encompasses a wide variety of manifestations occurring in due to presence of ectopic viable endometrial tissue in thorax. Various manifestations of TES include, pneumothorax, haemoptysis, haemothorax, lung nodule/mass, catamenial chest pain, diaphragmatic hernia and pneumopericardium [1]. Pneumothorax, often catamenial pneumothorax is the commonest presentation of TES, however it's unilateral, particularly right sided in nearly 97% of cases [2]. Here we are presenting a case of woman of reproductive age group who presented with bilateral pneumothorax secondary to TES, which is one of the rarest presentation of endometriosis with only a handful of cases being reported in literature, with the most recent being reported in 2020 by Christopher S. Sampson and Kathleen White [3].

Case Report

A 38 year old female, known hypothyroid, presented to ER on 15th July 2024, with acute onset dyspnoea since 3 days, that was exponentially increased over past 24 hrs. At presentation, her vitals were stable, maintaining a saturation of 93% on room air. On examination, patient is tachypnoeic, chest retractions were present, there is bilaterally reduced air entry with bilateral hyper resonant notes present on percussion. Thus, on suspicion of bilateral pneumothorax, patient was immediately placed on high flow oxygenation and a chest x-ray was taken that confirmed bilateral pneumothorax. As irregular right lung border was seen on chest x ray {Figure-1}, a CT chest was done {Figure-2}, which showed bilateral pneumothorax, with loculated air pockets seen on right side along with right lung collapse.

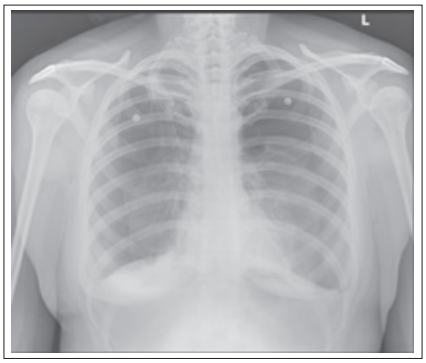

Figure 1

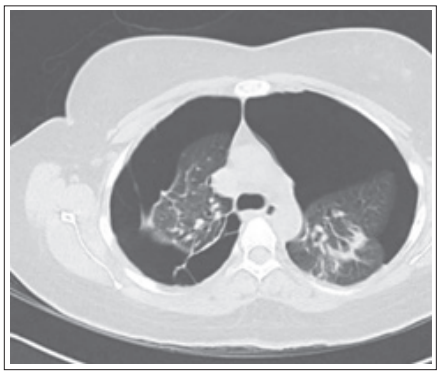

Figure 2

Patient was stabilised with bilateral 20F chest drain placement, initially on left side followed by right side, in the same sitting on the day of presentation. On further probing into patient's medical history post stabilisation with chest drains, it was revealed that patient's dyspnoea began with mild severity about a week back. It was noted that she had a history of air travel about 3 weeks ago. It was also noted that she had important obstetric and gynaecological history.

Even though she has regular cycles, she has history of dysmenorrhea with pain score of 4 out of 10, had history two IVF conceptions both delivered by caesarean section. Prior to her first pregnancy, she underwent laparoscopy for assessment of endometriosis. Her pap smear done 2 months back prior to presentation reported to be normal. Her last menstrual period was on 3rd July 2024 which was of regular duration with normal flow and dysmenorrhea. Prior to that it was on 29th May 2024 and she took norethisterone(5mg) for about 4 days to postpone the menstrual period for the month of June 2024 as she had plan for a religious trip.

Thus in view of this obstetric history, a gynaecologist was taken on board and USG abdomen with pelvis was done. It reported an endometrial thickness of 1mm, PCOS features in bilateral ovaries with slightly heterogenous echotexture of myometrium.

Post chest drain chest x rays and CT chest {figure-3} revealed a nearly completely expanded left lung, partially expanded right lung with right lower lobe consolidation. To rule out infective aetiology, bronchoscopy was done and bronchial washing collected

from right lower lobe and sent for microbiological and cytological examination, which were unremarkable.

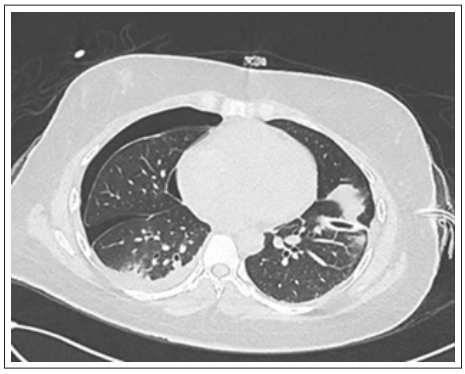

Figure 3

In view of non-expanding right lung and for aetiological assessment, patient was taken for bilateral VATS in the same sitting with cardiothoracic surgeon on board. Initially VATS was performed on the left side, a patch of endometriosis-like tissue was noted on dome of left hemidiaphragm {Figure-4}, it was resected and sent for histopathological examination. Along with a similar patch was noted on visceral pleura at the interlobar fissure {Figure-5}. Talc insufflation was done for pleurodesis, a 24F chest drain was placed, lung expansion was noted and the incision was closed.

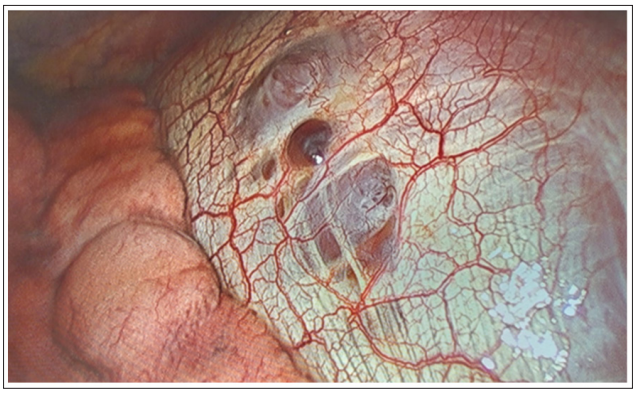

Figure 4

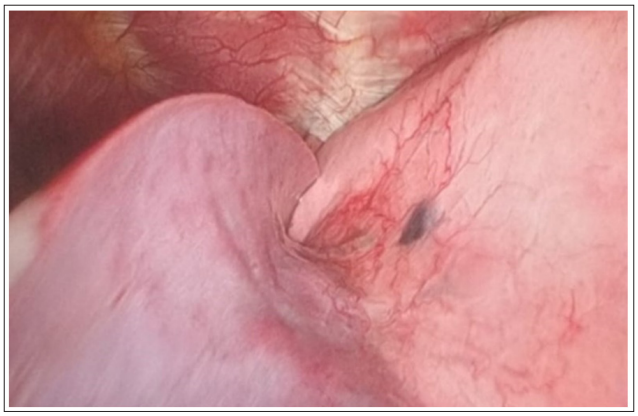

Figure 5

In the same sitting, right sided VATS was also performed, liver was noted to be herniating into thoracic cavity through a diaphragmatic rent {Figure-6}, liver was reduced into abdominal cavity and a mesh was placed to close the diaphragmatic rent. Talc insufflation was done for pleurodesis, a 24F chest drain was placed, lung expansion noted and the incision was closed. Procedure went uneventful, with a minimal blood loss.

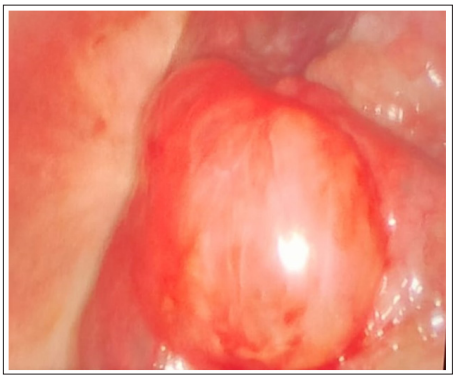

Figure 6

Following the procedure, patient was monitored via serial chest x rays, initially right sided chest drain was removed, followed by removal of left sided chest drain 3 days later. Histopathology confirmed thoracic endometriosis {Figure 7,8,9}.

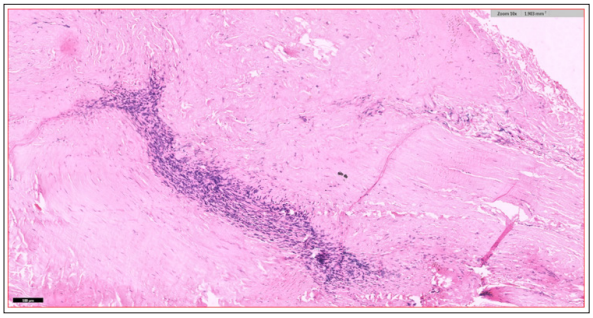

Figure 7: Shows Fibrocollagenous Tissue with Small Focus of Oval Darkly Stained Cells with Significant Crush Artefact. A Separate Irregular Gland is Noted which also show Cautery Artefact. Morphology of the Lining Epithelium can't be well Made Out. Focal Mesothelial Hyperplasia is Noted.

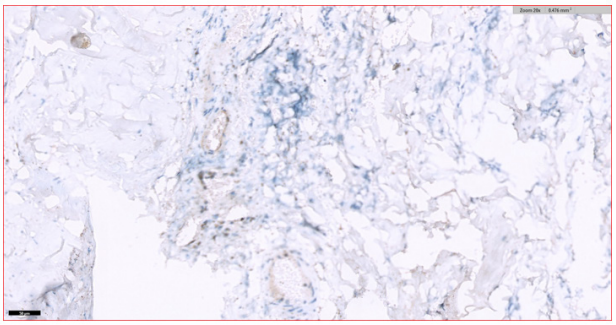

Figure 8: The Glandular Cells Are Positive for PAX8

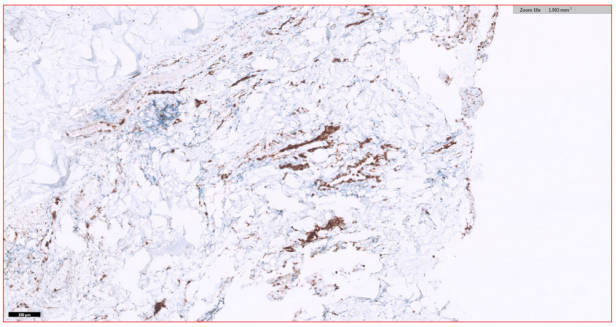

Figure 9: Stroma Showing CD-10 Positivity

Discussion

Endometriosis is a chronic oestrogen-dependent condition characterized by the ectopic implantation of functional tissue lining the uterus, i.e., endometrial glands and stroma outside of the uterine cavity, with the most common clinical symptoms being pelvic pain and infertility. Our case has history of infertility with both of her pregnancies being the result of IVF conception delivered via caesarean section.

The incidence of endometriosis is about 10% in females of reproductive age group [4]. In 49% of females of reproductive age having chronic pain, endometriosis is the underlying pathology [4]. Thoracic cavity is the most common extra abdominopelvic site for endometriosis, and presence of endometrial tissue in thoracic cavity results in wide spectrum of manifestations together called as Thoracic Endometriosis Syndrome (TES) [5].

The term Thoracic Endometriosis Syndrome (TES) was proposed by Joseph and Sahn in 1996. It includes catamenial pneumothorax, haemothorax, haemoptysis, lung nodules, chest pain, pneumomediastinum and pleural effusion. Out of these catamenial pneumothorax is the most common manifestation accounting up to 1/3rd of the cases of spontaneous pneumothorax occurring in woman of reproductive age group [6].

By definition catamenial pneumothorax is a spontaneous pneumothorax occurring in woman of reproductive age group 24 hrs before and up to 72 hours after the beginning of menstruation [7]. However, literature also indicates that it can occur as early 1 week before the beginning of menses and also can occur as late as 1 week after the last day of menses following a regular cycle [8].

Most of the cases in the literature have been unilateral, with 85-95% being exclusively right sided, with left sided being rare and bilateral being rarest [2]. Thus, in the clinical scoring system developed by Haga et al., to differentiate between spontaneous and a catamenial pneumothorax, the side of the pneumothorax – with right side carrying highest odds ratio, has been included in one of the 4 clinical variables [9]. The other 3 being – history of pelvic endometriosis, age and smoking history.

Our patient deviates from the scoring system in terms that she presented with bilateral spontaneous pneumothorax with no smoking history, but meets the age criteria and with positive pelvic endometriosis history.

Various Theories have been Proposed for the Pathophysiology of TES

1. The earliest and most accepted theory is Sampson's theory of retrograde menstruation and implantation. It is defined as the spillage of viable endometrial tissue from uterus into the pelvic cavity [10].
2. Anatomical asymmetry in the abdominopelvic cavity and the existence of peritoneal currents are some of the other likely explanations for the preponderance of right-sided thoracic endometriosis [10,11].
3. Lymphovascular spread and transdiaphragmatic spread through the diaphragmatic defects [12,13].
4. Microembolisation following trauma or uterine procedures [14,2].

Implanted endometrium might not always undergo cyclic hormonal changes like normal endometrial tissue [12]. However, as shown by the in vitro studies, certain growth factors like epidermal growth factor, insulin-like growth factor, and macrophage-derived growth factor may cause the proliferation of ectopic endometrial tissue, in addition to oestrogen [4,12]. Therefore, the ectopic endometrial lesions in the presence of a complex hormonal and proinflammatory environment tend to continue proliferation and angiogenesis [4].

Table 1: Shows Incidence of Some of The Common Manifestations of TES [13].

Clinical Manifestation	% of cases contributing to TES
Pneumothorax	72%
Haemoptysis	14%
Haemorrhagic pleural effusion or Haemothorax	12%
Lung nodules/mass	2%
Catamenial chest pain	NA
Hydropneumothorax	NA
Diaphragmatic hernia	NA
Pneumopericardium [15]	NA

Table 2: Showing Categories of Spontaneous Pneumothorax in Woman of Reproductive Age Group [8].

Catamenial and TE-related pneumothorax	Pneumothorax occurring in temporal relationship with menses with evidence of TE.
Catamenial but non TE-related pneumothorax	Pneumothorax occurring in temporal relationship with menses but without evidence of TE.
Non catamenial but TE-related pneumothorax	No temporal relationship of pneumothorax with menses but with evidence of TE
Idiopathic pneumothorax	Neither temporal relationship with TE nor there is evidence of TE.

Joseph et al reported that 97.5% catamenial pneumothorax cases occur on right side [2]. However, our case, though doesn't fit strictly into the definition of catamenial as it didn't align with the time period of either 24 hours before or 72 after the beginning of menses, there is evidence of thoracic endometriosis confirmed by histopathology from the tissue samples sent following VATS. {Figure of left sided endometrial tissue – on dome of diaphragm and at interlobar fissure; Presenting CXR}

Another important thing to note is that our case had bilateral pneumothorax with the evidence of endometriosis present in both the right and left thoracic cavity as confirmed by histopathology of tissue samples that we sent following bilateral VATS performed in a single session. Thus, our case falls into the rarest category of bilateral non catamenial but TE-related pneumothorax.

Roussett-Jablosnki et al. reported that up to one of the, every, four woman presenting with catamenial pneumothorax was having recurrent chest pain or scapular pain during their menses and nearly half of them had a history of obstetric or gynaecological procedure [14].

Even though our patient has undergone laparoscopy prior to her first pregnancy for assessment of endometriosis along with caesarean section for both of her IVF conceptions, patient never had any history of chest pain or scapular or shoulder pain during her menses.

History of obstetric or gynaecological procedure adds weight behind the theory of microembolisation leading to vascular spread of endometriosis [15]. Ottolina et al. noted that catamenial pneumothorax is associated with pelvic endometriosis, but not with infertility, which is a quite contrast to our case where the patient has pelvic endometriosis and infertility requiring IVF conceptions, with associated thoracic endometriosis [6].

Bobbio et al. reported that 2/3rd of women of reproductive age presenting with diaphragmatic rupture who were subjected to VATS, found to have endometriosis [16]. This is similar to our case where we have found liver herniating into the right thoracic cavity through a diaphragmatic rent while performing VATS on the right side which corelated with subtle change in the contour of right hemidiaphragm that was noted in the PA CXR and only in the coronal section of CT chest that were done upon the presentation, however the subsequent chest x rays done prior to taking the patient for VATS didn't show the same finding of right hemidiaphragm {Figure of liver herniation, Presenting CT chest and post ICD CXR}. The occurrence of diaphragmatic rupture greater than 6 months after undergoing VATS for catamenial pneumothorax could be attributed to TE rather than a complication of surgery [17].

The most common location for the diaphragmatic endometrial implants is the posterosuperior aspect of right hemidiaphragm, as the endometrial cells traverse through the paracolic gutter [18]. The phrenocolic and falciform ligament are expected to prevent the endometrial cells to get implanted on left hemidiaphragm, but our case proved to be an exception for this logic as we noted a patch of endometrial tissue on the dome of left hemidiaphragm, which was resected and was confirmed as endometrial tissue by histopathology [19].

In the literature, it has been described that, doing MRI chest seems to be more sensitive than CT chest during the menstruation period, particularly for woman presenting with catamenial chest symptoms, because MRI seems to pick up the diaphragmatic and pleural implants even when the CT chest seems to be non-diagnostic [20,21].

As such there is no specific laboratory test to detect TES. CA-125 levels seem to be elevated in TES as compared to normal [22]. Elevated CA-125 can predict recurrence with high specificity but with poor sensitivity [22]. A woman of reproductive age presenting with spontaneous pneumothorax with high levels CA-125 should always raise the suspicion of TES [22].

Bulen SE. in 2009 reported that gene expression profiling of endometrium can distinguish women with and without endometriosis [23]. In woman with endometriosis, endometrial cells circulate irrespective of development of TES or catamenial pneumothorax [24].

In 2020, Kiss et al. used gene expression profiling for analysis of genetic signatures associated with circulating endometrial cells (CEC) in woman with catamenial pneumothorax [25]. This revealed two genotypes of CECs, one associated with transdiaphragmatic communication between abdomen and thorax, other associated with pleural implantation of endometrial tissue [25]. Both these CECs express significantly higher HER2 as compared with CECs in woman with endometriosis but didn't experience a pneumothorax [25]. Their data shows a potential dose response relationship, as huge number of CECs were found in woman with recurrent catamenial pneumothorax [25].

Basing on this, Kiss et al. proposed assessing CEC gene expression profiles as a potential diagnostic test in young woman presenting with spontaneous pneumothorax to look for endometriosis [25]. This test also has the potential to identify the woman with endometriosis at high risk of developing catamenial pneumothorax [23,26,27].

VATS remains the method of choice as it serves as diagnostic as well as therapeutic modality, and also it can deal with a spectrum of TES pathologies like diaphragmatic defects, diaphragmatic implants, visceral and parietal pleural implants, parenchymal implants, blebs, and bullae [1].

Although nearly half of the catamenial pneumothorax patients who underwent VATS were found to have diaphragmatic defect, there was not statistical association between presence of a diaphragmatic defect and development of catamenial pneumothorax [1]. It was also noted that those people who had visceral and parietal pleural implants found in VATS were at 5 times higher risk for haemoptysis [1]. In our case we noted a visceral pleural implant in the left lung at the interlobar fissure, however patient never had any episode of haemoptysis.

Currently there was not much literature available on role of medical thoracoscopy in TES, barring few case reports like that of Ravi Kanth Velagapudi & John P. Egan III article published in 2021, where they used medical thoracoscopy in a patient with known abdominopelvic endometriosis presented with recurrent haemothorax suspected of having pleural endometriosis. Here they have opted for medical thoracoscopy instead of VATS as the patient requested a lesser invasive alternative to VATS [28].

Bronchoscopy has extremely limited and specific role in TES. In a case of catamenial haemoptysis, bronchoscopic airway examination can locate the anatomical region that is bleeding which can be followed by VATS wedge resection or endobronchial laser ablation [29-31]. Performing bronchoscopy during menses increases the success of locating endobronchial endometrial tissue [32].

Hormonal therapy alone has high recurrence rate once discontinued; thus, anti-gonadotropin agents are used both pre- and post-operatively [33]. Recommended duration for an anti-gonadotropin agent is a minimum of 6 to 12 months with regular follow ups with a multidisciplinary team [6]. The anti-gonadotrophic agents include cyclic or continuous oral contraceptives, dienogest, danazol, cyproterone acetate, and GnRH agonist like leuprolide [34,35].

As such there are no comparative studies between cyclical and continuous oral contraceptives, one study showed that cyclic oral contraceptives had higher recurrence rates compared with continuous oral contraceptives and GnRH agonists [16].

Conclusion

Thoracic Endometriosis Syndrome encompasses a wide variety of thoracic issues resulting secondary to endometriosis, with pneumothorax being the most common manifestation of it. Although the pneumothorax associated with TES is most commonly unilateral, particularly right sided; unilateral left sided involvement is rare and a presentation with bilateral pneumothorax as seen in our case is one of the rarest. Often the TES-pneumothorax is associated with diaphragmatic defects, as noted in our case too with liver herniating into right thoracic cavity via diaphragmatic rent, but there is no statistical association between the two.

VATS remains the modality of choice as it is both diagnostic and therapeutic, as well as allowing for uniform pleurodesis with talc insufflation, thus playing a major role in significantly reducing recurrence. However, the best way for the treatment seems to be combination of hormonal therapy of minimum of 6-12 months duration along with VATS intervention, preferably hormonal therapy started preoperatively and continued post operatively. Thoracic Endometriosis Syndrome remains an enigmatic entity to screen in woman with known abdominopelvic endometriosis, however, the latest research by Kiss et al. in genetic expression profiling of circulating endometrial cells (CECs) seems to have the potential to develop into both screening as well as diagnostic test of choice [25].

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