

## Catamenial Pneumothorax: A Unique Case with Multiple Sites of Endometriosis and a Villar's Nodule

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**Introduction:** Endometriosis is the presence of endometrial tissue, glands, and stroma outside the uterine cavity. It affects 10 to 15% of women and it is estrogen-dependent, thus remains closely associated with the perimenstrual period, i.e., 72 hours before or after the menstrual cycle. Various hypotheses were postulated, but the etiology and pathogenesis remain obscure and difficult to explain. However, retrograde outflow of menstrual blood with the open fallopian tube is the most popular explanation. The thoracic Endometriosis is a rare entity, but catamenial pneumothorax has the maximum share in incidence of >72% and is one of the alarming respiratory emergencies. The present case is unique with Endometriosis involving multiple sites.

**Case Report:** A 30-year-old fertile, lean, thin (Low BMI) woman presented with a history of sudden onset of shortness of breathing with dark umbilical nodule and rectal bleeding during the menstrual cycle. The catamenial pneumothorax was managed with conservative treatment, and the hormonal therapy was initiated with favourable results. The rectal bleeding was managed with a stool softener. USG whole abdomen and Doppler study of the umbilical region depicted and confirmed the diagnosis of umbilical Endometriosis (Villar's Nodule) with minimal ascites.

**Conclusion:** Endometriosis is a chronic, benign disease that affects women throughout the world. In most occasions, it remained confined to the Pelvis with vague symptoms including backache. A detailed menstrual history should be taken and is an integral part of all medical examinations. This case becomes unique and worth presenting with umbilical Endometriosis, rectal bleed, hemorrhagic ovarian cyst, together with catamenial pneumothorax as a rare emergency of Respiratory Medicine.

### Introduction

Endometriosis is the presence of endometrial tissue, glands, and stroma outside the uterine cavity with accompanying inflammation. Smolarz, B. et al.,<sup>1</sup> mentioned Endometriosis as a "mysterious" disease, and its exact cause is not known; however, J.A. Sampson first time in 1927 gave the term 'endometriosis' and hypothesized that (Sampson's Theory) the retrograde menstrual blood with exfoliative mucosal cells is likely to be transported and implanted in the Pelvis. Similarly, the other contiguity organs, like the abdomen, rectal, bowel, cutaneous, etc.,<sup>2-4</sup> are some examples, though rarely migration of cells through diaphragmatic fenestration, they enter the thorax, thus the pleura and or lung parenchyma involvement is also reported.<sup>2,5-7</sup> The transport and implant action by blood and lymphatic routes are also possible; however, it is worth mentioning

that etiology and pathogenesis are still difficult to explain.<sup>1</sup> The ectopic endometrium also behaves in a similar way during periodic cyclic menstrual hormonal changes as with eutopic endometrium. It is estrogen dependent; thus, in most cases, the symptoms remained perimenstrual, i.e., 72 hours before and after menstrual bleeding, and occur in women between menarche and menopause. Endometriosis affects 10-15 % of women, and most of the cases remain précised to the Pelvis with chronic backache, and a high of 30-45 % was reported in infertility cases.<sup>1,3,4</sup> Endometriosis is much more common with previous gynecological procedures, e.g., laparoscopy, laparotomy, D&C, cesarean section, etc.<sup>2</sup> However, the majority of cases remain asymptomatic or undiagnosed and under-notified/ reported.

The Pelvis is the most common site for Endometriosis, and the literature mentions that 80% of females have open fallopian tubes with a chance of retrograde outflow

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of menstrual blood; however, only a few of them suffer from Endometriosis. Ovarian Endometriosis occurs in the form of a superficial lesion or as an endometrial cyst that USG can easily diagnose<sup>1</sup> The other pelvic sites are the uterine ligament, the Fallopian tube, etc. The usual symptoms of pelvic Endometriosis are chronic and vague, including chronic pelvic pain, dysmenorrhea, deep dyspareunia, dysuria, dyschezia, fatigue, infertility, irregular menstruation, blood in the stool, diarrhea, or constipation. These symptoms perhaps affect physical, mental, sexual, and social well-being; thus, a strong clinical suspicion enables us to consider it in a differential diagnosis.<sup>3,4</sup> Endometriosis at multiple sites, involved with pneumothorax, is worth reporting, as happened in our case.

### Case Report

A 30 years young female had presented with chest and lower abdominal pain and breathlessness for 2 days. On examination was found to have a BP100/60 mm Hg, Pulse 88/minute, SpO<sub>2</sub>96%, RR 18/minute. She was having a lean built with a BMI of 18.6 kg/m<sup>2</sup>. Her chest X-ray (Figure 1) depicted a right-sided pneumothorax. The patient refused initially admission but came back after 3 days and was admitted with additional complaints of Ghabrahat and anal bleeding since 2 days. A pigmented nodule was observed at the navel (Figure 2). Her CBC and urine reports were essentially normal except for low Hb 8.7% (normochromic normocytic), blood group B+ with adequate platelets on smear, and the liver and renal functions were within normal limits. USG abdomen reported no organomegaly, a grossly normal-sized uterus with endometrial thickening of 7 mm, the left ovary was obscured, and the right ovary had a 3.3x3 cm hemorrhagic cyst and mild ascites. The umbilical region was reported with a 1.8x1.3 cm-sized central echogenic component and significant vascularity at the navel region for which a Doppler study was performed (Figures 3 and 4). The surgeon and gastroenterologist advised for sigmoidoscopy, but the patient flatly refused. The patient was negative for HBsAg, HIV, and HCV. No history of Surgery, HT, DM, or blood transfusion. Obstetric history revealed one full-term normal delivery 15 years back (alive male) and one induced abortion (of 3 months of pregnancy) 3 years back, not followed by D & C. Menstrual history: menarche at 12 years of age, the normal period of 4-5 days/ 28-30 regular, dysmenorrhea. Two months back she had, she had a similar menstrual pain in the chest and lower abdomen, and was diagnosed and registered as extra-pulmonary tubercular pleural

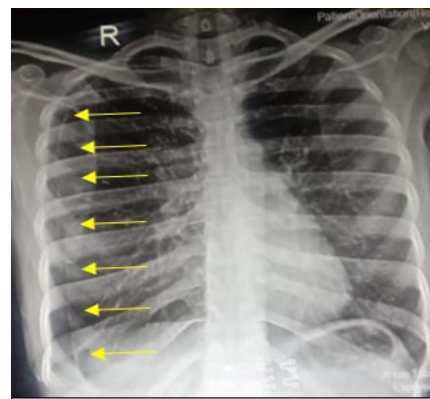


Figure 1: Arrows shows right sided pneumothorax



Figure 2: Umbilical endometriosis (Villar's node)



Figure 3: USG of umbilical swelling

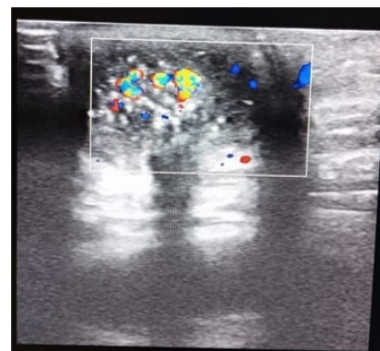


Figure 4: Doppler of umbilical swelling

effusion on the right side. Diagnostic pleurocentesis reported exudative and hemorrhagic aspirate with glucose 56 mg%, protein 4.2 gm%, LDH 981 U/L, and ADA 31 U/L, microscopic cell counts 220/cumm, predominant 80% lymphocytes, mesothelial cells 20%, admixed with RBCs, and no atypical or malignant cells seen.

The pneumothorax in the present study was managed conservatively with high-flow intermittent oxygen, antibiotics, and supportive antispasmodic, antacid, vitamins, and haematinics. A minimal rectal bleed was managed with counseling/assurance, and with a stool softener and laxative. Our patient was treated with hormonal therapy in consultation with a Gynecologist. The pneumothorax remarkably improved, and she was discharged after 5 days of hospitalization. The follow-up of the patient remained asymptomatic on the four consecutive menstrual cycles.

## Discussion

Endometriosis is not uncommon, with reported prevalence being 10-15 percent among fertile females, with maximum incidence developing in the Pelvis. The backache, too, is a common complaint in females, perhaps due to a subtle, small endometrial bleed with repeated inflammation and organization, and that might lead to it being a chronic benign disease. Smolarz, B et al., elaborated on certain predisposing factors for Endometriosis, including, e.g., early menarche before 11years, low BMI, low birth weight, age between 25-29, a small number of births, and more often diagnosed in infertile women.<sup>1</sup> There are a lot of hypotheses postulated to define the involvement of various organs or systems; however, the symptoms depend upon the site and size of the ectopic tissue. The Extra pelvic Endometriosis involves the cutaneous, peritoneum, gastrointestinal tract, including the rectum, etc.<sup>3,4</sup> Similarly, R Panicker et al mention Villar's Nodule, first described in 1886, is an extremely rare incidence, being only 0.1-0.5 % among external Endometriosis.<sup>8</sup> Our case report belongs to a unique presentation having Endometriosis at different sites, including e.g., hemorrhagic ovarian cyst, umbilical (Villar's Nodule), rectal bleed, and pleuro-pulmonary as CP.

The thoracic cavity is the most common location for extra-abdominal and pelvic Endometriosis.<sup>6</sup> Tomasz Marjański et al in their review article mentioned the diaphragmatic theory of air passage, migratory, and or the microembolism metastatic theories to explain the development of thoracic foci of Endometriosis. They further mentioned that the recurrent pneumothorax

associated with menses was described by Maurer et al. in 1958, but Lillington et al was credited for defining catamenial pneumothorax (CP) in 1972.<sup>2</sup> CP is a very rare ailment and only 3% to 6% of cases among spontaneous pneumothorax meet the definition. Among them, the CP is an alarming and life-threatening emergency, because it leads to sudden lung collapse at any time during MC, and the extent of (minimal to tension pneumothorax) collapse cannot be predicted. A typical presentation of CP is unilateral in 80-95% of cases, and that too on the right side<sup>2</sup> has also been observed in our case, but rarely could it be on the left due to lymphatic or hematogenous spread. It is ideal to manage and consider chemical/ mechanical pleurodesis in cases of recurrent CP with or without repair of diaphragmatic fenestration with a modality like VATs,<sup>9</sup> and Arbat, A P et al., has mentioned bronchial artery embolization for catamenial hemoptysis.<sup>5</sup> Our case has been managed conservatively along with hormonal therapy. However, a Meta-analysis of thoracic Endometriosis (TE) concluded that among them catamenial pneumothorax had the highest incidence at 72% followed by hemoptysis at 14%, pleural hematomas in 12%, and lung nodules in only 2% of cases.<sup>2</sup> The catamenial pneumothorax occurs perimenstrual thus the recurrence is most likely, and that may require pleurodesis to create adhesion between visceral and parietal pleura to obliterate the pleural space.

Smolarz B et al<sup>1</sup> have mentioned Pharmacological, surgical, or combination treatment, i.e., Physiotherapy in Endometriosis to improve the pelvic floor. Somehow, it is not always practical to remove or excise the endometrial tissue from different sites, either for a diagnostic or therapeutic purpose. The absence of these histopathologic findings does not rule out TE.<sup>6</sup> A repeated symptom or bleed during MC should be considered as a clinically diagnosed endometriosis. So far, no specific marker for Endometriosis has been identified but imaging modalities like X-ray, USG, contrast CT or MRI serve the purpose of supportive evidence as demonstrated in our case.

## Conclusion

Endometriosis is a chronic benign disease of a fertile woman. On most occasions, it remained concise to the Pelvis with vague symptoms including backache, dysmenorrhea, dysuria, dyschezia, and infertility. A detailed menstrual history should be considered as an essential and integral part of medical examination. It may the diagnosis of some of the rare and emergency events like catamenial pneumothorax. A tissue diagnosis is not

always possible for multiple-site endometriosis; hence, clinical diagnosis with supportive evidence should be considered. The absence of these histopathologic findings does not rule out TE.

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