

Uncommon cause of recurrent spontaneous pneumothorax in a young female

Rajoor UG^{*1}, Seema BN²

¹Associate professor, Department of Medicine, ²Assistant Professor, Department of OBG, SDM college of medical sciences and hospital, Sattur, Dharwad-580009, state-Karnataka, India.

*Corresponding Author:

Name: Dr. Umesh. G. Rajoor

Email: drumeshrajoor@gmail.com

Abstract: Recurrent spontaneous pneumothorax associated with the menstrual cycle or catamenial pneumothorax. Here we presented such uncommon recurrent spontaneous pneumothorax in a young a case report of 22-year-old, woman. She had symptoms of right-sided chest pain, shortness of breath and cough with scanty sputum associated with menstruation. In conclusion, Clinical suspicion and recognition of the temporal relationship of the patient's symptoms with menses are essential to establish the diagnosis. Radiographic studies and thoracoscopy may support the diagnosis. Pathologic evidence of thoracic endometrial tissue is not present universally. Therapeutic interventions, which include medical and surgical options, must be individualized for each patient.

Keywords: pneumothorax, menstrual cycle, diagnosis, young woman

INTRODUCTION

Recurrent spontaneous pneumothorax associated with the menstrual cycle [1] or catamenial pneumothorax is an extremely rare entity of which the etiology still remains obscure. Although numerous theories have been postulated, the lack of consistency from one patient to another has precluded the formulation of a single, unique mechanism. Management still remains controversial [2]. We report a case of a catamenial pneumothorax as cause of recurrent spontaneous pneumothorax in a young female.

CASE REPORT

A 22-year-old, non-smoking woman was admitted with complaints of sudden onset of right-sided chest pain, shortness of breath and cough with scanty sputum associated with menstruation for 3 days duration in march 2010. The same symptoms had occurred in 2001, 2002 and 2005 during almost every menstrual cycle. In 2001, she was diagnosed to have pneumothorax following onset of menstruation, which was managed with intercostal tube drainage (ICD) (fig 1). In 2002 and 2005 she had similar presentation and managed with ICD (fig 2 & 3). She also had no evidence of a possible familial spontaneous pneumothorax.

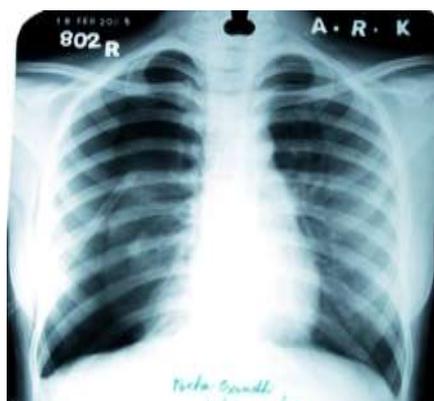


Fig 1 right pneumothorax with ICD in 2001

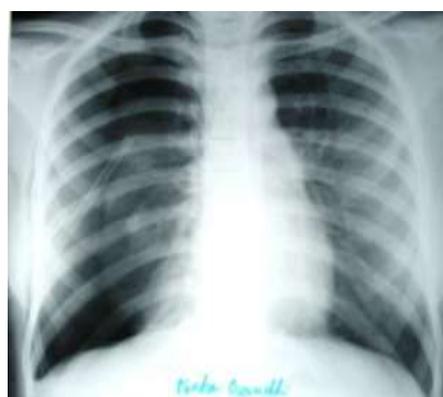


Fig 2 right pneumothorax with ICD in 2002



Fig 3 right pneumothorax with ICD in 2005



Fig 4 right pneumothorax in 2010

On physical examination, decreased breath sounds and hyper resonance were notable on the upper zone of the right hemi thorax. Laboratory data were within the normal limits. Chest X-ray showed collapse of the right lung resulting from pneumothorax (fig 4). Computed tomography showed pneumothorax but neither evidence of any bullous lesion nor any finding consistent with endometriosis. The patient refused any attempt of an invasive procedure but a thoracentesis, which confirmed the pneumothorax and helped to evacuate the trapped air in the pleural space. She was discharged 4 days later following clinical and radiological re expansion of her lungs.

DISCUSSION

Endometriosis is the growth and function of endometrial tissue outside of the uterine cavity [3]. Endometriosis predominantly affects women of reproductive age group and is estimated to affect approximately 5-10% of the female population [4]. The most accepted explanation for endometriosis is metastatic implantation theory [5]. This theory states that there is retrograde transport of endometrial tissue into the peritoneal cavity. Thorax is a rare site of endometriosis. It can present in various manifestations, the most common of them is catamenial pneumothorax [6]. Other common manifestations include catamenial haemoptysis, catamenial haemothorax, lung nodules, isolated catamenial chest pain and catamenial pneumo

mediastinum. Collectively, these entities have been termed the “thoracic endometrial syndrome.” It is suggested that cyclic changes in endometrial tissue in the thorax may be responsible for the clinical manifestations. Confirmatory pathologic evidence of thoracic endometrial tissue is not identified universally. The diagnostic hallmark is the onset of symptoms within 48 hours of the onset of menstruation. Rarely, pneumothorax development precedes menstruation. Symptoms are related temporally with menstruation but often do not occur with every menstrual cycle [7]. Catamenial pneumothorax accounts for 2.8–5.6% of all episodes of spontaneous pneumothorax in women [8]. There is an overwhelming right-sided predilection for all manifestations of thoracic endometriosis. Pleural lesions are almost exclusively right-sided, whereas lung lesions have no such predilection [3]. The possible cause for this right sided predilection could be explained from the fact that the lymphatic drainage does not take place evenly over the whole diaphragmatic surface, but is more extensive on the right side [9]. So the embolized endometrial tissue is more likely to be carried to the right than the left. Pathogenesis of thoracic endometriosis is debated on two theories: microembolization theory and peritoneal-pleural migration. Microembolization theory states that endometrial tissue can be transported through the lymphatics or vascular channels into the lung parenchyma. Peritoneal-pleural migration theory states that endometrial tissue reaches the pleura through the peritoneum via defects in the diaphragm [10].

The diagnosis of thoracic endometriosis syndrome requires an appreciation of the entity and subsequent recognition of the temporal pattern of the patient’s symptoms. It is not uncommon for patients to have numerous cyclic episodes before the association with menses is recognized and the diagnosis is established. A temporal relationship with menses should be sought in any woman of childbearing age who presents with an unexplained pneumothorax. Symptoms may not occur with every menstrual cycle, which further contributes to the challenge of this diagnosis [11].

Pelvic ultrasound is the first and foremost investigation done in case of suspected endometriosis [12]. In thoracic endometriosis it assumes even more importance due to the fact that pulmonary endometriosis usually coexists with pelvic endometriosis. High resolution computer tomography (HRCT) is the modality of choice for localization of endometrial deposits in the lung and pleura. Magnetic resolution imaging (MRI) is considered superior to CT in diagnosis of endometriosis due to the presence of blood products in the endometrial deposits.

Treatment of catamenial pneumothorax is controversial. It has been suggested that the management of a catamenial pneumothorax is identical to that of a pneumothorax of any etiology¹. Medical line of therapy is the first line of treatment. Drugs aimed at suppressing ectopic endometrium – oral contraceptive pill, danazol, gonadotropin releasing hormone (GnRH), synthetic progestins.

Surgical treatment [2]: Medical thoracoscopy with talc poudrage, video-assisted thoracoscopic surgery (VATS), chemical sclerosis/ pleurodesis, pleural abrasion, partial pleurectomy, diaphragmatic repair with polymesh, thoracotomy, stapling of blebs, bullectomy.

CONCLUSION

Catamenial pneumothorax is a rare entity with varied clinical manifestations. Clinical suspicion and recognition of the temporal relationship of the patient's symptoms with menses are essential to establish the diagnosis. Radiographic studies and thoracoscopy may support the diagnosis. Pathologic evidence of thoracic endometrial tissue is not present universally. Therapeutic interventions, which include medical and surgical options, must be individualized for each patient.

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