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THORACIC ENDOMETRIOSIS SYNDROME IN A 75-YEAR-OLD WOMAN

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ABSTRACT

Thoracic endometriosis syndrome (TES) is a rare condition where endometrial tissue is found in the pleura, parenchyma, and/or diaphragm. Haemoptysis is a common manifestation of pulmonary endometriosis and can be cyclic, aligning with the menstrual cycle. Patients can present with residual cough once menses have stopped. Other manifestations are recurrent pneumothorax and/or pleural effusions associated with menses. On imaging, pulmonary nodules or implants of endometrial tissue along the diaphragm can be present and be confirmed by biopsy. While most cases of TES occur in menstruating females in their thirties on the right side, our case is unique since we report a postmenopausal female with TES on the left side without the typical clinical manifestations.

KEYWORDS

Thoracic endometriosis syndrome, pleural mass, post-menopausal female

LEARNING POINTS

- Physician awareness about thoracic endometriosis appearing as a lung mass on the left side in a postmenopausal woman is essential to aid in prompt diagnosis and management of this rare pathology.

INTRODUCTION

Thoracic endometriosis (TES) is a rare form of extra pelvic endometriosis affecting predominantly women of reproductive age. It can manifest in a variety of symptoms such as haemoptysis, dyspnoea, and chest pain that is typically associated with the menstrual period. Clinical manifestations can develop as early as 72 hours after onset of menstruation with recurrent catamenial pneumothorax accounting for 73% of cases[1]. Much less common are pulmonary nodules which can appear as a lung mass on imaging due to the fibrotic tissue surrounding the implanted endometrial tissue. While TES is highest in menstruating females ages 30-35 years, our current patient was postmenopausal with no history of endometriosis or ever being on oestrogen therapy. Diagnosis can be challenging due to atypical presentations and underdiagnosis as symptoms are not immediately connected to endometriosis in this demographic. Untreated thoracic endometriosis can lead to several complications such as recurrent pneumothorax and haemothorax, ultimately causing respiratory distress. A high

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degree of physician awareness is required for early diagnosis and treatment of thoracic endometriosis.

CASE DESCRIPTION

A 75-year-old female with a past medical history significant for smoking, hypertension, coronary artery disease status, diabetes mellitus type 2 and osteoporosis presented to the pulmonary clinic to establish care. The patient denied any chest pain, shortness of breath, cough, night sweats, weight loss or abdominal pain. She stopped smoking 20 years ago, reached menopause several years ago and denied any history of hormone therapy. On physical examination, the patient had good air entry bilaterally with no rales, rhonchi, and wheeze. She had negative egophony, and vocal/tactile fremitus, however, she did have increased anterior-posterior diameter and increased expiratory phase. Her recent pulmonary function tests showed forced expiratory volume (FEV1) of 1.43 l, 81%, forced vital capacity (FVC) 1.93 l, 82%, and ratio 0.76. Her laboratory parameters are shown in *Table 1*. A computed tomography (CT) scan of the chest performed 3 years prior to presentation showed no acute pathology *(Fig. 1)*. She underwent low-dose CT scan of the chest without contrast for screening of lung cancer and was found to have a 3.1 x 2.8 cm left lower lobe lobulated mass with adjacent mild pleural thickening and some comet tailing *(Fig. 2)*. No enlarged lymph nodes were noted. A positron emission tomography (PET) scan was done which showed moderately hypermetabolic mass at the left lower lobe of the lung. No hypermetabolic thoracic lymph nodes. No hypermetabolic lesions within the neck, abdomen, or pelvis. The decision was made to get a biopsy due to high suspicion of malignancy. The patient underwent CT-guided core needle biopsy of left lung mass. The pathology report showed benign endometrial glands with benign endometrial type stroma. The immunostains confirmed diffuse PX-8, ER, and CD10 immunopositivity. The case was discussed in detail with the patient. Since she was asymptomatic at the time, a decision was made to follow clinically.

Parameter	Result
Sodium (mEq/l)	139
Potassium (mEq/l)	5.2
BUN (mg/dl)	17
Creatinine (mg/dl)	1.07
AST (U/l)	27
ALT (U/l)	15
Alkaline phosphatase (U/l)	91
Haemoglobin (g/dl)	9.4
Platelets (cells/µl)	184
WBCs (cells/µl)	5.90

Abbreviations: BUN, blood urea nitrogen; AST, aspartate aminotransferase; ALT, alanine aminotransferase; WBC, white blood cells.

Table 1. Laboratory parameters.

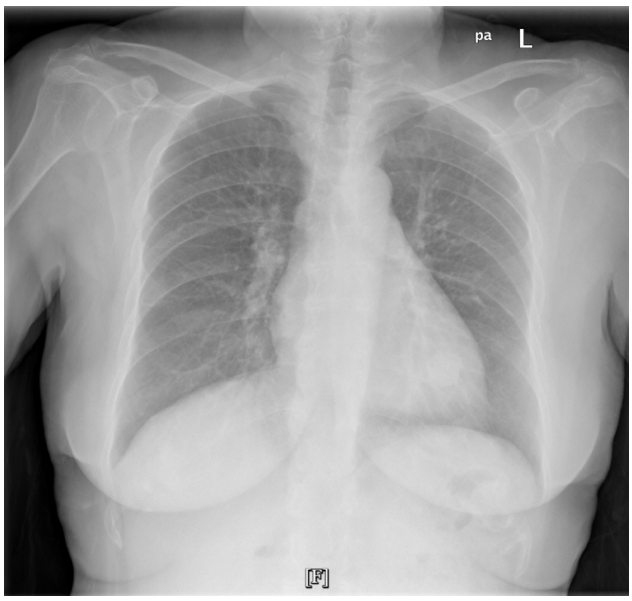


Figure 1. Chest X-ray taken three years earlier showed no acute pathology.

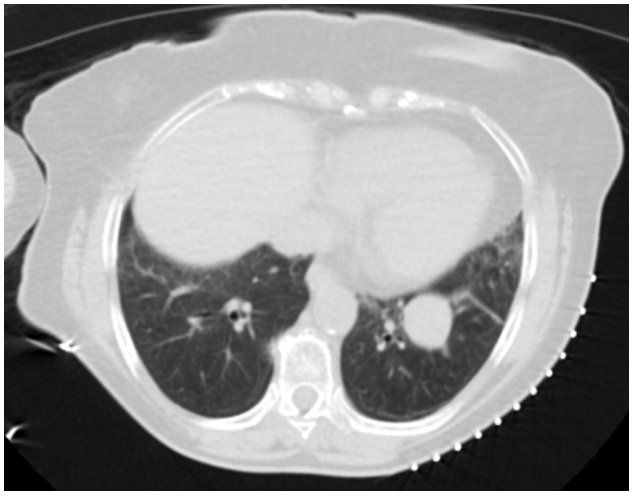


Figure 2. CT of the chest shows a 3.1 x 2.8 cm lobulated mass in the left lower lobe, with adjacent mild pleural thickening and some comet tailing. No enlarged lymph nodes are noted.

DISCUSSION

Endometriosis is the presence of normal endometrial mucosa outside the uterine cavity[2]. It is usually confined to the pelvic organs particularly the uterosacral ligament, the ovaries and the broad ligament but it can also be found in extra-pelvic organs including abdomen, thorax and skin[2]. Endometriosis is a common gynaecological disease in women of reproductive age group since it is oestrogen dependent and indicates active ovarian function[3]. It is extremely rare for this disorder to occur in postmenopausal women and has a prevalence of 2% in this population[3]. The presence of endometrial glands and stroma in the lung parenchyma,

airway and pleura is called thoracic endometriosis syndrome (TES)[4]. It is a rare disorder with over 110 documented cases since it was first described in 1956[5]. Moreover 92% of the TES cases occur in the right hemithorax. We report a unique case of left sided TES in a postmenopausal woman.

Thoracic endometriosis in a post-menopausal female is a rare condition typically occurring in those who have undergone hormone replacement therapy (HRT) or have residual endometrial tissue that remains hormonally active. Thoracic endometriosis can present 1-2 days before menses in women of childbearing age and has been reported in the literature to present with haemoptysis in 7% of the population, lung nodule in 6%, pneumothorax in 73% of the women and haemothorax in 14% of population[4]. In our case the patient was asymptomatic with an incidental finding of lung nodule on the left side found during screening for lung cancer. In post-menopausal women, these symptoms may not follow a cyclic pattern due to lack of menstrual cycles but can be triggered by exogenous oestrogen.

The pathogenesis of TES is still elusive, and several hypotheses have been proposed to explain the presence of intrathoracic endometrial implants. One theory is coelomic metaplasia, another suggests retrograde menstruation which involves migration of the endometrial tissue through the transperitoneal and transdiaphragmatic approach and the third theory hematogenous or lymphatic embolization from the uterus[4,5]. In postmenopausal women oestrogen threshold theory is proposed which suggests that when a certain level of oestrogen is exceeded in postmenopausal women a transient focus of endometriosis is activated[5].

The diagnosis of TES is challenging and is often delayed especially if a temporal relationship with menses is not identified. The presence of the aforementioned symptoms in a post-menopausal female with history of HRT or previous endometriosis should raise suspicion for thoracic endometriosis and initiation of applicable diagnostic imaging and histopathological confirmation. Chest X-ray is non-specific whereas ultrasound plays a chief role in the diagnosis since TES may be associated with abdomen and pelvic endometriosis. CT scan is poorly specific but is the preferred imaging modality which may show ground glass infiltrates, nodular lesions or hypo attenuated areas of endometrial implants[4]. Magnetic resonance imaging (MRI) is superior to CT scan and may demonstrate hyperintense lesions of endometriosis[5]. Pleural fluid cytology is rarely helpful, and bronchoscopy utilization is limited since TES mostly occurs in the periphery. Histopathological examination can be used in some cases to confirm the diagnosis[5]. In our case a chest CT scan showed a left lower lobe nodule, and the diagnosis of TES was confirmed with histopathological analysis. The effective treatment strategies are controversial due to lack of appropriate therapeutic trials however medical, surgical or combination approaches can be employed. Medical therapy is focused on suppression of oestrogen secretion but has a higher recurrence rate[4]. The definitive treatment of TES remains hysterectomy with bilateral salpingo-oophorectomy in women who are not on hormone replacement therapy. Video-assisted thoracoscopic surgery (VATS) or open thoracotomy can also be considered if surgical treatment is suggested[2].

CONCLUSION

Thoracic endometriosis is an underrecognized and complex condition. Physician awareness of this rare pathology is required for early diagnosis and appropriate management. A high index of suspicion is required in order to make the diagnosis especially in post-menopausal women.

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