Case Report

Thoracic endometriosis syndrome with bloody pleural effusion in a 28 year old woman

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Abstract

Thoracic endometriosis, rarely encountered, is characterized with the localization of functional endometrium tissue in pleura, lung parenchyma or tracheobronchial system. A 28 years old female patient visited our clinic with complaints of cough and shortness of breath for the last two months. Right-sided massive pleural effusion was detected in the chest radiography and thorax computed tomography. Exudative fluid was aspirated with a haemorrhagic appearance on thoracentesis. Cytology was evaluated as suspicious. "Signet ring cells" were reported in pleural biopsy. Diagnostic biopsy was performed by video-assisted thoracic surgery (VATS) on the patient whose fiber-optic bronchoscopy was normal. The histopathological diagnosis was reported as "pleural endometriosis". Chemical pleurodesis was applied with asbestos-free chalk. The thorax HRCT (high resolution computed tomography) performed during menstruation was normal. Thoracentesis was needed 3 times for recurrent pleural effusion in the follow-ups and then parietal pleurectomy was performed. The patient is in postoperative 10th month follow-up and evaluated as normal clinically and as radiologically.

Pleural endometriosis should be considered as a differential diagnosis in female patients with infertility with chest symptoms. Video-assisted thoracoscopic surgery can be useful in the diagnosis and treatment of these patients and chemical pleurodesis and parietal pleurectomy should be considered among the treatment options.

Keywords: Thoracic endometriosis, Pleural effusion, Thoracic surgery.

Introduction

Endometriosis is the presence of functional endometrium in an organ or tissue rather than uterus. It is generally in ovary, uterosacral ligament and "cul-de-sac". While its incidence in the general population is not known accurately, endometriosis is diagnosed in 30 % of the women that are researched due to infertility and 40-50% of the women who are evaluated for dysmenorrhoea or pelvic pain.[1]

Thoracic endometriosis (TE) is a situation that is rarely encountered. The functional endometrium tissue locates in the lung parenchyma, pleura or airways. This tissue responds to hormonal changes in association with the menstrual cycle. TE has symptoms and findings emerging as catamenial pneumothorax, haemothorax, haemopneumothorax, haemoptysis, and pulmonary nodules.[2]

Diagnosis of TE may not be possible with non-invasive methods. We present the case of a 28 years old female who had ovarian malignancy mimicking pleura metastasis and diagnosed as pleura endometriosis with video-assisted thoracic surgery (VATS) and who was applied parietal pleurectomy with thoracotomy in her follow-ups as it was a fairly rare case.

Case Report

Complaints of cough, shortness of breath and chest pain was present for two months in a 28 years old female patient. She had been infertile for 6 years. Her menarche was in her 13th year. She complained of painful menustrual cycles. She had been operated due to ovarian cyst rupture 6 years ago. Post-operative pathology was evaluated as corpus haemorrhagicum cyst. Her haemogram and biochemical examinations were normal. CA-125:90,9 U/ml. Breathing sounds were decreased in the right hemithorax. Pleural fluid was observed in a significant amount on the right side in postero-anterior chest radiograph. Massive free pleural fluid was present from apex to base in the right hemithorax and passive atelectasia was observed in the neighbouring lung in thorax computed tomography (Figure-1,2). (Posteroanterior chest X rays demonstrated a significant right pleural effusion. Thorax computed tomography scan revealed pleural effusion with loss of lung volume.) The pleura fluid obtained after thoracocentesis was exudative and had a haemorrhagic appearance. Pleura fluid cytology was reported as "signet ring-like atypical cells"

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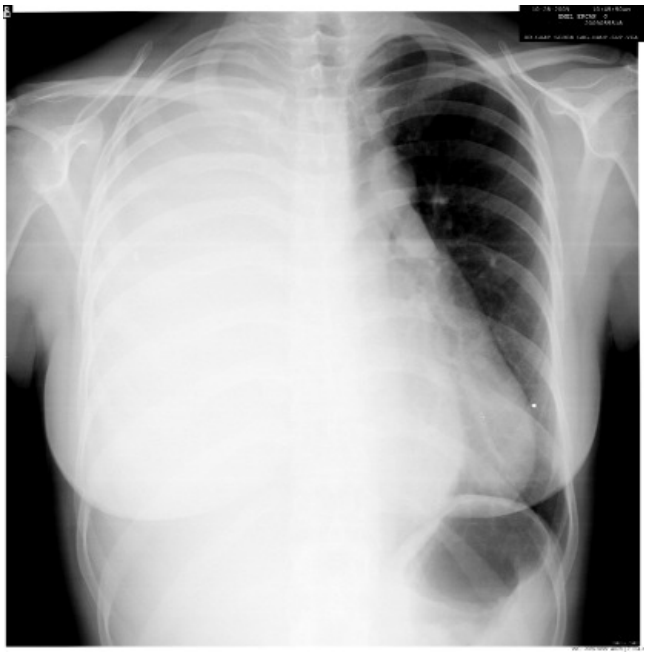

Figure-1: Posteroanterior chest X rays demonstrated a significant right pleural effusion.

Figure-2: Thorax computed tomography scan revealed pleural effusion with loss of lung volume.

and percutaneous pleura biopsy was reported as "benign". The screening examination of the aobdomen revealed ascites and a mass with a diameter of 5 cm (cystic lesion with dense ingredients) in left ovary. Fiber-optic bronchoscopy was normal. FVC:1.87 (%48), FEV1:1.52 (%45), FEV1/FVC: 97. VATS was planned for diagnosis for the patient who had "dyspnoea, massive pleural fluid on right side and ovarian mass and acid". About 2500cc haemorrhagic fluid was aspirated. Millimetric colour changes which are dark brown and in patches joint and scattered were seen in the chest wall and the diaphragmatic pleura. Pleural biopsy was taken from this section. Postoperative histopathological result of the biopsy materials was reported as "pleural endometriosis". Thorax HRCT was performed for controlling the case during her menstrual cycle which started on the 2nd post-operative day. Lung parenchyma was evaluated as normal. Chemical pleurodesis was applied with asbestos-free chalk from the operation drain. The patient was also seen by the gynaecologist and oral contraceptive was given for treating endometriosis. Laparoscopic cystectomy was performed and the histopathological diagnosis revealed endometrioma. Thoracentesis was performed 3 times for recurrent pleural effusion in right hemithorax in her 1.5 year follow-up. Total parietal pleurectomy was decided as the pleural fluid recurred. The second operation was done with VATS and 3000cc haemorrhagic fluid was aspirated. Total pleural pleurectomy was done. The patient is now in 10th months follow-up and is clinically and radiologically normal. Her Ca-125 value was 28,8 U/ml on the last visit.

Discussion

Endometriosis is the localization of active endometrium tissue generally in ovaries and peritoneal serosa rather than uterus. It can localize in cervix, vagina, vulva and abdominal wall in decreasing frequency. Extragenital endometriosis is a rarely encountered situation. TE is classified as pleural and parenchymal types. While pleural type is seen more frequently at the rate of 83%, its prevalence as intraparenchymal or bronchial is 17%. TE composes four different clinical tables as pneumothorax, haemothorax, haemoptysis and pulmonary nodule according to the tissue in which it is predominant.[3-5] In a research made with the results taken from the data of 110 cases diagnosed as TE and/or of the series, pneumothorax was diagnosed in 73%, haemothorax in 14%, haemoptysis in 7% and nodule in 6% cases. Pleural type endometriosis seen in right hemithorax in 85%, causes pneumothorax or pleural effusion and with symptoms of dyspnoea and chest pain.[6]

There are different theories for the formation of thoracic endometriosis. In the pleural type thoracic endometriosis; local metaplasia of cholemic epithelium (metaplasia theory), trans diaphragmatic pass of endometrium tissue

via abdomen and diaphragmatic fenestrations from uterus and fallopian tubes (retrograde menstruation) and its implantation into the thorax cavity has been given as reasons. In parenchymal type thoracic endometriosis; haematogenous expansion of endometrium tissue after surgical operations such as curettage and caesarean and microembolization with a mechanism similar to pulmonary embolism was held responsible.[7,8]

Anamnesis has an important place in the diagnosis of the disease. The infertility and pelvic surgery history of our case was the most important clue indicating thoracic endometriosis in differential diagnosis and the pathologist was informed. It is important for the differential diagnosis that the radiological findings are nonspecific and symptoms vary with menstrual cycle. Cytologic examination of the pleural fluid is rarely useful. The benefit of bronchoscopy is also limited as lesions are peripherally localized in parenchymal type TE. The pleural fluid cytology was non-diagnostic and bronchoscopy was normal in our case. As in our case, diagnostic surgery with VATS or thoracotomy is necessary in more than 60% of the cases.[7]

Medical approach in endometriosis treatment is focused on suppressing endometrium tissue by blocking the estrogen activity. For this purpose, gonadotropin-releasing hormone (GnRH) and danazol are the therapeutic agents used. Recurrence rate is more than 50 % for TE. The high rate of recurrence is possibly due to the insufficient suppression of thoracic endometrium tissue and embolization's repeating from pelvic focus.[9] As hysterectomy and bilateral salpingo-oopherectomy requires estrogen replacement treatment in women in reproductive age, it can cause the reactivation of thoracic endometrium tissue. In the cases that are complicated with haemothorax, pneumothorax and pleural effusion, chemical pleurodesis is a treatment option that is recommended before more aggressive surgical procedure. VATS is a reliable and minimally invasive surgical method for both diagnosis and treatment. By this technique, bleeding lung tissue can be seen and partial resection, pleural abrasion or partial pleurectomy can be applied on pigmented lesions.[6-10] We also observed disseminated brown lesions on the chest wall of our case by video thorachoscopy. No additional surgical operation was made as the final histopathological diagnosis was presented after the first VATS operation. However total parietal pleurectomy was applied by a second operation due to recurrent pleurisy in spite of medical treatment during the follow-ups.

Consequently; thoracic endometriosis should be considered for differential diagnosis for haemothorax, pneumothorax and haemoptysis etiology in the female patients who have infertility and pelvic surgical operation history. VATS operation is a reliable and minimal invasive technique for the histopathologic diagnosis and treatment of the disease. We believe that parietal pleurectomy is effective besides medical treatment in the prevention of recurrent pleural effusion in these patients.

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