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Catamenial Haemothorax Masquerading Pulmonary Tuberculosis? A Case Report of a 28 Year Old Woman who Presented with Recurrent Haemothorax and Empyema Thoracis

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Abstract Catamenial haemothorax is a clinical manifestation of endometriosis of the pleural cavity. This is a case report of a 28 year old woman who presented with recurrent catamenial haemothorax who had histological confirmation of pelvic endometriosis and uterine leiomyoma. A presumptive diagnosis of catamenial haemothorax was made as it was not possible to get a histological confirmation of endometriosis of the pleural cavity. She was treated effectively with gonadotropin-releasing hormone GnRH agonist-leuprorelin. Antituberculosis treatment was however added when she developed empyema thoracis instead of haemothorax at a time she was experiencing amenorrhoea. Pulmonary tuberculosis being an endemic disease remained the main differential diagnosis but was never confirmed at any stage by investigations conducted. She has since not experienced any recurrence in eighteen months of follow-up after treatment was completed. **Conclusion:** Pulmonary or thoracic endometriosis though rare; should be considered seriously in women of reproductive age who present with recurrent catamenial haemothorax. Due to obscured locations of the endometriosis lesions in the thoracic cavity; the diagnosis may be difficult to confirm since access to investigative tools is limited in developing countries.

Keywords: *catamenial haemothorax, pulmonary endometriosis, thoracic endometriosis syndrome, extra pelvic endometriosis*

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1. Introduction

Endometriosis is the presence of functional endometrial tissue[endometrial glands and stroma outside the uterine cavity [1,2,3], most common locations are dependant parts of the female pelvis such as the ovaries, fallopian tubes, uterosacral ligaments, sigmoid colon and pelvic peritoneum [1,2,3]. Some extra pelvic locations of endometriosis include the umbilicus, abdominal scars, kidneys, pleural cavity, lungs, transverse colon, breasts, lymph nodes, nasal mucosa, the arms and legs [1,2,3]. The true incidence of endometriosis in unknown, however it is estimated to be between 5% to 15% of women in their reproductive years [1,2,3]. There are several theories to explain the histogenesis of endometriosis but the most popular theory is that endometriosis results from retrograde menstruation [2,3]. In contrast to the theory of retrograde menstruation, is the theory that endometriosis arises from metaplasia of coelomic epithelium or proliferation of embryonic rest and the theory of the endometrium being transplanted via the vascular and lymphatic systems in rare and remote location [2,4,5].

In thoracic endometriosis, there is functioning endometrial tissue in the pleural cavity, lung parenchyma, airways and diaphragm [1,2]. It may manifest as *catamenial pneumothorax, catamenial haemoptysis, catamenial haemothorax, catamenial haemopnuemothorax and lung nodules* [2,3,5] depending on the location on the endometriosis lesions within the thoracic cavity. The true incidence of thoracic endometriosis is not known however 75% of thoracic endometriosis cases reported present as catamenial pneumothorax whereas catamenial haemoptysis and catamenial haemothorax represent 8.3% of cases [3,6]. It is estimated that about a third of women hospitalized for spontaneous pneumothorax are actually experiencing catamenial pneumothorax [3,7], endometriosis of the lungs or pulmonary endometriosis. Catamenial haemothorax is the usual clinical manifestation of endometriosis affecting the pleural cavity [3,8,9].

Catamenial haemothorax presents as haemorrhagic pleural effusion together with non specific symptoms such as pleuric chest pains, cough and shortness of breath

which may be occurring periodically with the menstruation[2]. Most of the cases previously reported were diagnosed based on the clinical history of the patient as histological confirmation of ectopic endometriosis is not always done [1]. Investigations such as chest x-rays, CT, MRT, bronchoscopy may be useful in diagnosis of conditions in the chest but have limitations and inconsistent findings [1,2].

The treatment of endometriosis can be medical, surgical or both [2,3]. Medical treatment is usually the first line of management of thoracic endometriosis usuing GnRH antagonist such as danazol, hormonal agents such as progestatins, oral contraceptive pills, and GnRH analogues [2,3]. The use of these drugs for medical treatment is aimed at induction of amenorrhea or suppression of ovarian function. Recurrent bleeding in the ectopic implants is one of the most important pathophysiological processes to interrupt [3] for an improvement to be noticed. Patients presenting with catamenial pneumothorax, catamenial haemopnuemothorax or catamenial haemothorax as in the case reported would initially require surgical intervention such as thoracocentesis and chest tube placement as first-step in the emergency room until further action is taken [3]. After performing the emergency lifesaving thoracocentesis and chest tube placement, when the patient's condition becomes stable, the option of medical treatment can then be initiated. Hysterectomy and bilateral salpingo-oophorectomy is the treatment of last resort when other options have failed [3], this should be reversed for those in the later ages of the reproductive years and do not want to have any more children.

Our experience in the management of this unusual case of recurrent catamenial haemothorax is being reported so that attention is drawn to thoracic endometriosis when it presents as a clinical entity.

2. Case Report

A 28 year old nulliparous woman was referred for gynaecological consultation with a diagnosis of uterine leiomyomata and suspected thoracic endometriosis in early 2013 due to recurrent haemothorax. She presented five months earlier to the hospital with fever, chill, anorexia and a productive cough which lasted about one mouth. She was treated with broad spectrum antibiotics for right lobar pneumonia after investigations that included sputum negative smear and chest X-rays. Subsequently she developed episodes of haemothorax occurring periodically with her menstruations. She was treated with broad spectrum oral antibiotics on three occasions after varying amounts of 1-1.5 litres of haemorrhagic pleural effusion was drained; but sputum smears and laboratory studies of the effusion remained negative for malignancy, acid fast bacilli [AFB] and other organisms. Chest x-rays however confirmed right sided pleural effusion on all three occasions before thoracocentesis and placement of chest tube was performed. She experience monthly recurrence of haemothorax during her menses and also lost up to 12kg of weight during 5-6 months duration of illness but she did not have recurrent fever, persistent cough or haemoptysis. As the diagnosis of pulmonary tuberculosis could not be confirmed, she was referred for gynaecologist and cardiothoracic specialist consultations and investigation on suspicion of thoracic endometriosis or malignancy.

She did not have any family history of significance and there was no positive history of contact with tuberculosis. She was singled, lived alone in an apartment and did not smoke tobacco or drink alcohol. Her menarche was at 11 years, she had a menstrual cycle length of 24-26 days and bleeds for 7 days. She had positive history of dysmenorrhea and chronic pelvic pain of three years duration, and had episodes right sided chest pain which radiated to her shoulders during her menses which was managed on analgesics a few months before the monthly recurrences of haemothorax. She did not experience vaginal discharge, intermenstrual bleeding, or dyspareunia.

On examination, she was calm and well looking, body mass index of 21.1kg/m2 with no pallor, jaundiced or fever. The thyroid gland and both breasts were normal with no peripheral lymphadenopathy. Her respiratory was 20 cycles per minute with reduced air entry on the right middle and lower lobes with dull percussion note. Air entry was adequate on the left lung with vesicular breath sounds and no crepitation. Other findings including the cardiovascular and gastrointestinal systems, liver, both kidneys and the spleen were all normal. There was a multi-nodular mass in the lower abdomen arising from the pelvis about 26 week's size gestational uterus. The mass was not tender and there was sign of free fluid in the abdomen. The vulva and vagina were normal and the cervix was closed and posterior. The adnexae and pouch of Douglas were normal and there was no blood or vaginal discharge noticed.

Summary of results of some investigations done during the period before her referral included normal haematological investigations, negative sputum for Acid Fast Bacilli[AFB], routine examination and culture of pleural aspirate were all negative for malignancies and infections. She however had ultrasound reports confirming leimyoma of the uterus. Multiple chest x-rays and CT-scan showing previous episodes of right sided haemothorax Figure 1a, Figure 1b, Figure 2a and Figure 3a and repeat chest x-rays showing normal findings after thoracosentesis and chest tube drainage as in Figure 2b and Figure 3b below.

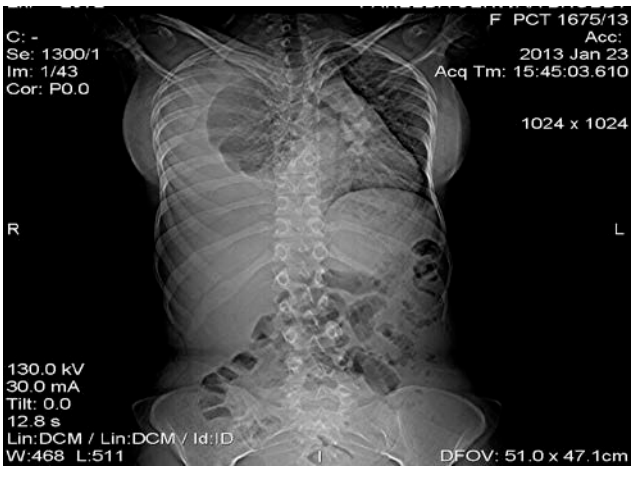

Figure 1a. CT scan of the chest and abdomen showing massive right pleural effusion

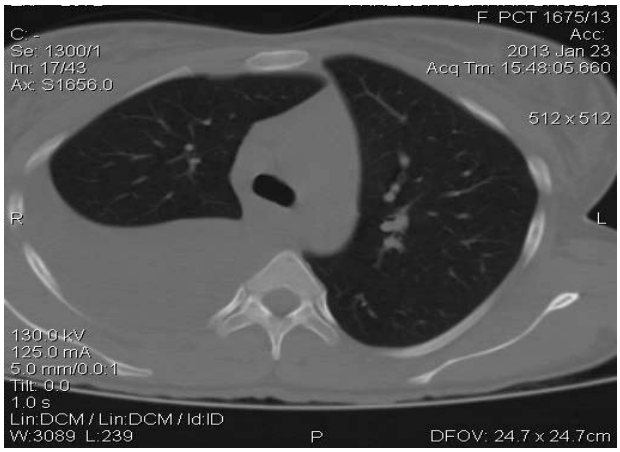

Figure 1b. Section of CT scan of the chests showing massive right pleural effusion and normal left lungs

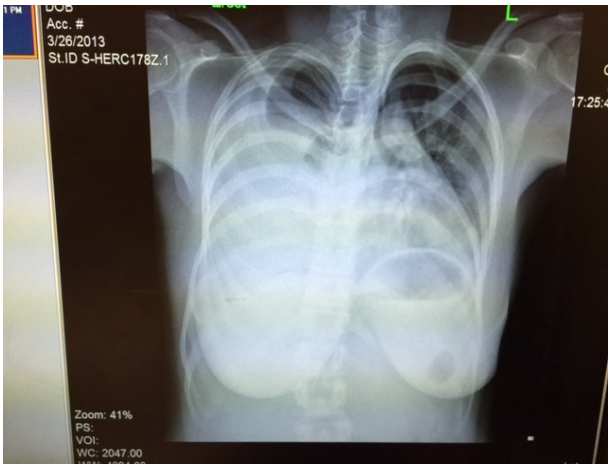

Figure 2a. Chest X-ray showing massive right pleural effusion. March 2013

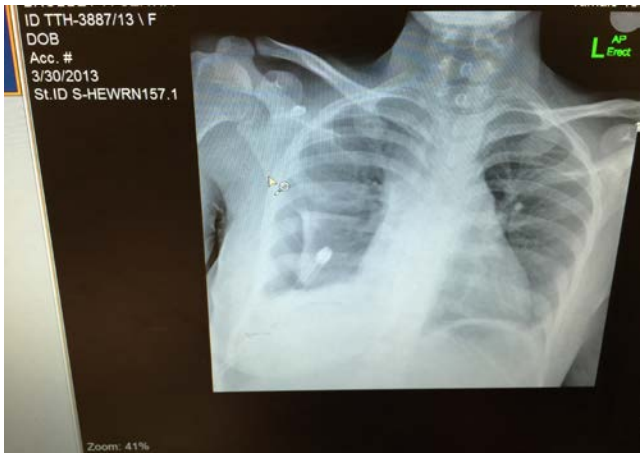

Figure 2b. Chest X-ray showing drainage of right effusion 4 days after chest tube placement

After the gynaecological and cardiothoracic surgical specialist consultation, the need for additional investigations including a repeat of CT-Scan or MRI of the chest, and possibly tissue biopsies of any lesions within the pleural cavity for histological studies were discussed with her. She was not prepared financially for these additional investigations when she experienced a recurrent episode of haemothorax with severe respiratory embarrassment on the last day of her menses in May 2013. She was admitted to the intensive care unit where thoracostomy was performed and chest tube was retained for five days after chest x-rays confirmed the recurrence of right haemothorax with left side remaining normal. An ultrasound scan done showed multiple uterine myomas with some significant amount of free fluid in the abdomen which was not obvious from clinical examination.

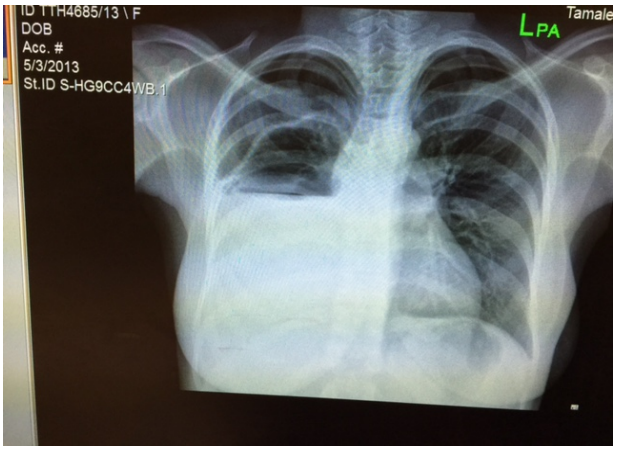

Figure 3a. Chest X-ray showing massive right pleural effusion. May 2013

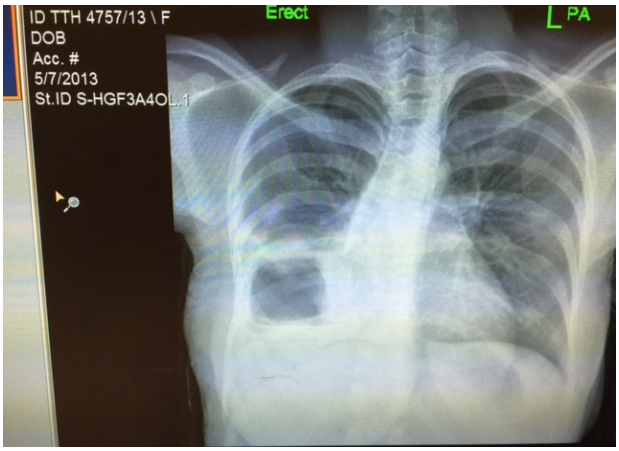

Figure 3b. Chest X-ray showing drainage of right effusion after chest tube placement

She was given prophylactic parenteral broad spectrum antibiotics and analgesics for 72hrs and was transfused three pints of whole blood when haemoglobin was noticed to be 8.0g/dl and was symptomatic for anaemia. Other laboratory investigations done included serological test for human immunodeficiency virus, repeat routine examinations and cultures of pleural effusion were negative for malignancies, AFB and other bacterial infections. The pleural effusion was completely drained and exploratory laparotomy and drainage of the free fluid in the abdomen was performed. Finding at laparotomy was some 180-200mls of chocolate brown colored fluid which was drained. Other significant finding included the multi nodular uterine myomas and severe pelvic endometriosis confirmed by histopathology of biopsies of tissues taken from different sites within the pelvis.

The multi-displinary team of specialist upon discussion agreed on a presumptive diagnosis of thoracic endometriosis with catamenial pneumothorax and the need to commerce an effective treatment using GnRH analogues [leuprorelin].

The treatment of GnRH analogues injections [leuprorelin] were given monthly for three months. Three weeks after comment of the GnRH analogues treatment she developed empyema pleurae instead of haemothorax. She had thoracostomy and insertion of drainage tube which drained about 1200mls of purulent effusion, laboratory investigations conducted on the purulent pleural aspirate still did not confirm pulmonary tuberculosis; antituberculosis treatment was however also initiated as a precautionary measure. She had myomectomy successfully performed five months after commencing treatment with GnRH analogues when the size of the uterine leiomyomas regressed to about 16 weeks gestation. The adhesions in the pelvis had also regressed with disappearance of much of endometriosis lesions seen during the earlier laparotomy. Her menses resumed about six weeks after the last GnRH analogue injection; it was heavy and lasted for five days and was treated with tranexamic acid. She has since had regular menstruation established without dysmenorrhea or recurrence of haemothorax and she is also not experiencing chronic pelvic pains either during more than eighteen months of follow-up.

3. Discussion

This is a case of recurrent catamenial haemothorax due to thoracic endometriosis in a 28 year old nulliparous woman was managed successfully with GnRH agonist [leuprorelin]. The presumptive diagnosis of thoracic endometriosis was based on recurrent catamenial haemothorax; a condition which was not confirmed by histology since tissue could not be taken from the pleural cavity. Results of cytological studies of the effusion were not diagnostic of endometriosis, malignancy or pulmonary tuberculosis.

A history of recurrent catamenial heamoptysis or catamenial haemothorax when pulmonary tuberculosis cannot be confirmed should draw attention for the diagnosis of pulmonary or thoracic endometriosis to be considered. The diagnosis of extra-pelvic endometriosis may be difficult or delayed[2] as in this case reported due to the rare nature of the condition or the patient's symptoms in relation to the menstrual cycle may not be apparent [1]. Pulmonary endometriosis or thoracic endometriosis usually presents in women of the reproductive age group with a mean age of 34 years with over 90% of the lesions located within the right hemithorax and 14% presenting as catamenial haemothorax [2,3,7,12]. Some 50-84% of women with pulmonary or thoracic endometriosis have a history of pelvic endometriosis with the symptoms of pelvic endometriosis preceding some 5-7 years [3,7,12]. Though histological confirmation of thoracic endometriosis was not obtained, there was however histological confirmation of pelvic endometriosis in this same case. The clinical symptoms of catamenial haemothorax and effectiveness of treatment with GnRH analogue provides basis for the correctness of the presumptive diagnosis. Treatment with GnRH analogue provided dual advantage of shrinking the large uterine myomas and suppressing symptoms of pelvic endometriosis making the myomectomy procedure easier for the large myomas which presented as an additional pathology.

Imaging studies or bronchoscopy during menses and repeated during the midcycle may assist in the diagnosis of pleural or bronchopulmonary disease. The disappearance of the previous findings strengthens the clinical suspicion [13]. The C T findings for pulmonary endometriosis may include well-defined opacities, nodular lesions, thin-wall cavities, or bullous formations, transient radiologic densities in the affected part of the lung [1,14]. Chest X-rays were done during the management of this case and it only provided value in diagnosis of the pleural effusion and its drainage but not its cause.

Although antituberculosis treatment was added when she developed empyema thoracis instead of haemothorax at a time she was experiencing amenorrhoea, the cause of the empyema may be from bacteria infection due to the thoracocentesis and insertion of drainage tube on several occasions as the diagnosis of pulmonary tuberculosis was still not confirmed by any of the investigations that were conducted on the purulent pleural aspirate. Since thoracic endometriosis is not common, the diagnosis may be delayed or missed as the symptoms are not specific. Pulmonary tuberculosis may be the primary focus of investigations and pose a differential diagnostic challenge in tuberculosis endemic parts of the world when thoracic endometriosis presents as catamenial hemoptysis as in a case reported by Yuz et al [11] or catamenial haemoptysis as in this case report It is important a presumptive diagnosis be made based on symptoms and recurrence of periodic haemothorax at the time of menstruation so that treatment options are considered early.

Patients with thoracic endometriosis usually undergo either surgery or medical treatments with GnRH agonists considered more effective in controlling recurrences of catamenial pneumothorax, particularly when used for prolonged periods of up to 1 year [3,15,16] though no large-scale randomized trial has been conducted and the optimal treatment regimen still remains controversial [1,4,16].

4. Conclusion

Pulmonary or thoracic endometriosis though rare; should be considered seriously in women of reproductive age who present with recurrent catamenial haemothorax. Due to obscured locations of the endometriosis lesions in the thoracic cavity; the diagnosis may be difficult to confirm since access to investigative tools is limited in developing countries where other diseases such as pulmonary tuberculosis could masquerade the condition areas where it is endemic.

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