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Case Report Open Access

Unusual Cause of Recurrent Chest Pain: Catamenial Pneumothorax

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Introduction

Catamenial pneumothorax (CP) is a condition of collapsed lung occurring in conjunction with menstrual periods. Typically, it occurs in women aged 30-40 years, but has been diagnosed in young girls as early as 10 years of age and post-menopausal women. Some sources claim this entity represents 3-6% of pneumothorax in women [1]. In regard of the low incidence of primary spontaneous pneumothorax (i.e. not due to surgical trauma etc.) in women (about 1/100'000/year). It is the most frequent manifestation of Thoracic endometriosis, characterized, in the majority of the cases, by right side localization. The resection of suspected areas of visceral or parietal pleural endometriosis. Thoracic endometriosis has been considered a rare clinical condition but it is probably underestimated in the literature. As well as partial resection of the diaphragm in the case of nodules and/or perforations, allows the histological diagnosis of endometriosis. Because of the high recurrence rate, treatment of catamenial pneumothorax should combine surgery and hormonal therapy.

We report, in this regard, a clinical case.

Case Report

We report the case of a 19-year old girl who consults for sudden right chest pain that occurs on the first day of the menstrual cycle. She reported regular menstrual cycles. But she complained of dysmenorrhea associated to ovarian endometriosis.

Physical examination showed tachycardia, normal respiratory rhythm, normal oxygen saturation but lack of vocal vibrations and breathe sounds in right side. Chest X-ray showed total right pneumothorax. A chest tube has been set up successfully (Figure 1). During the surgery, there was no macroscopic lesion objectified. The patient had no pleural biopsy.

One month later, she consults for the same pain. Chest X-ray showed partial right pneumothorax. The patient underwent a pleural chemical and mechanical refreshing. The patient was treated by Decapeptyl®.

Eight months after, the patient consults for sudden left chest pain that occurs on the middle of the menstrual cycle. The chest X-ray and tomography show a partial left pneumothorax (Figure 2). The patient underwent a pleural chemical and mechanical refreshing. By questioning the patient, she confessed that she stopped Decapeptyl® by herself. Then, it was retained catamenial pneumothorax because of his early menstrual cycle occurrence and frequent right location.

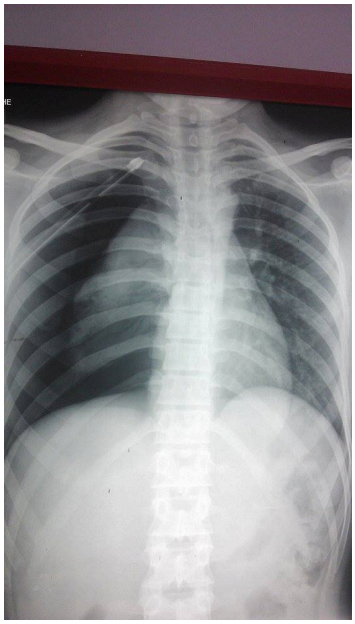

Figure 1: A Chest X-ray showing a right pneumothorax with a chest tube in place.

Discussion

Endometriosis is the condition in which the endometrial tissue is present outside the uterine cavity. It is most often found in the pelvic structures such as ovarian, uterine ligaments, pelvic peritoneum, cervix, lips and vagina.

Thoracic endometriosis syndrome (TES) is the presence of endometrial tissue in or around the lung.

There are 4 TES entities: catamenial pneumothorax (CP), catamenial hemothorax, hemoptysis, and pulmonary nodules. We are interested, in our observation, to CP. Endometriosis can affect up to 15% of women in their reproductive first year [1-3].

CP is an extremely rare entity, characterized by recurrent pneumothorax occurring between the day before and 72 hours after the start of the menstrual cycle, usually in the chest cavity of the right side in women on productive age [3,4].

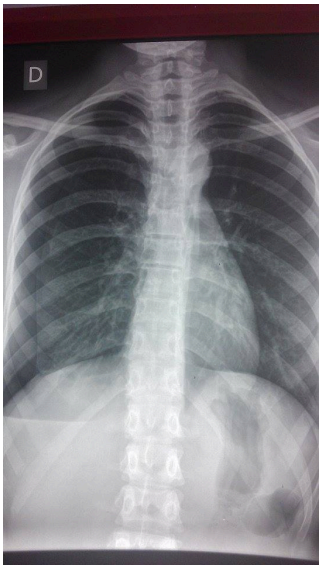

Figure 2: ChestX-ray and tomography showing a partial left pneumothorax.

In 1938, Schwartz described a woman with endometriosis inguinal node with hemoptysis with a "lung tumor" that bled in each menstrual cycle. Since then, endometriosis was documented in lungs, bronchi, pleura and diaphragm.

Over the past 100 years, several studies have focused on a better understanding of the prevalence, clinical manifestations, diagnosis and treatment of TES. The two largest retrospective studies of this period showed that the maximum impact of TES occurs between the ages of 30 and 35 and CP is the most common presentation of TES [5,6].

Several mechanisms to explain the CP have been proposed in the literature; however, no clear etiology was established.

It remains unclear how endometrial tissue migrates to the thoracic cavity. The theory of the presence of endometrial tissue in the lungs has been raised, including the coelomic metaplasia, or conversion of the peritoneal/pleural epithelium in an endometrial tissue under the influence of physiological stimuli [7]. This theory explains the presence of endometrial tissue patients without a uterus including men on prolonged estrogen therapy. However, it fails to explain the right-sided thorax predominance seen in most TES cases.

An additional theory is that endometrial transplantation occurs through lymphatic/vascular microembolization, explaining the presence of both intrapulmonary and other extra-uterine sites of implantation.

TES occurs almost exclusively in the right hemithorax (approximately 95% of cases). There are several potential reasons for this laterality. Physiologically, endometrial tissue located within the peritoneum likely follows the same directional flow, landing more commonly on the right diaphragm [6,8].

Finally, although the congenital diaphragmatic hernias are much more common on the left, the congenital diaphragmatic defects are known to occur more frequently on the right, which leads to the predominance of the right side of TES [9]. Your patient had two episodes on the right and two on the left.

According to the anatomical mechanism, there is an open connection during menstruation between the external atmosphere and the peritoneal cavity, due to the absence of the cervical mucus. Air can migrate through the fallopian tube into the abdominal cavity and diaphragmatic fenestrations in the chest, causing a pneumothorax. Bulla or bleb leads to pleural rupture [10].

In this context, Korom reported that in 73 patients with catamenial pneumothorax, 38.8% had diaphragmatic lesions, such as diaphragmatic endometriosis or perforation, 29.6% had endometriosis of the visceral pleura, 23.1% had bullae/blebs, and 8.5% had no lesions [11].

In addition, the change in the intraperitoneal pressure with breathing causes the right hemidiaphragm to contract against the liver, known as the "piston effect", which optionally allows the implantation of endometrial and/or migration through the diaphragm.

The absence of intrathoracic lesions is associated with the prostaglandin F2α tromethamine model [12]. High serum levels of prostaglandin F2α tromethamine during menstruation cause vasospasms and bronchospasms. Alveolar tissue damaged from vasospasms may possibly rupture if bronchospasms of any degree impede expiration. This hypothesis could explain the fact that we found no macroscopic lesions during surgical intervention in our case.

Imaging by tomography may assist in accurate diagnosis, showing the presence of pulmonary nodules in case of hemoptysis, cavity or scarring in patients with catamenial pneumothorax.

The gynecological investigation should be systematic. Their negativity does not exclude the diagnosis of endometriosis. In our case the diagnosis of ovarian endometriosis prior to the onset of the first episode of pneumothorax which facilitated the diagnosis of PC.

Both tomography and magnetic resonance imaging have been shown to be useful in the diagnosis of TES. Tomography imaging may reveal diaphragmatic endometrial implants as hypo-attenuating areas or identify single or multiple pulmonary nodules. Unfortunately, findings on tomography in patients with hemoptysis are nonspecific but may sometimes be represented as a focal ground- glass opacity or consolidation.

Endometrial implants on magnetic resonance imaging, however, will be hyper-intense, not hypo-attenuating as seen on tomography [13].

There is a limited role for bronchoscopy in diagnosis because most pathologic features are located in the peripheral lung.

Shiota et al were able to make the diagnosis of pulmonary endometriosis by cytologic examination of bronchial washings, although earlier studies indicated bronchoscopy was useful if performed within 1 to 2 days of the onset of menses [14].

Additionally, bronchoscopy may play a role in the localization of a bleeding lobe.

Video-assisted thorascopic surgery allows direct visualization of the lung and diaphragmatic surfaces, and descriptions of the findings include perforations on the surface of the diaphragm, brown and violet endometrial deposits, and larger masses [15,16]. Your patient underwent the surgery in the middle of the menstrual cycle. During the operation, we found no macroscopic lesions.

Treatment of spontaneous pneumothorax is much discussed in the literature. Some authors propose a thoracoscopic treatment from the first episode of primary spontaneous pneumothorax, while others only offer a surgical attitude in the second episode, after conservative treatment by the establishment of a chest tube. Thoracoscopy with wedge resection and apical pleural abrasion is currently accepted by most surgical teams as an initial surgical treatment instead of thoracotomy [17,18]

The management of catamenial pneumothorax differs slightly from the primary spontaneous pneumothorax. We offer a thoracoscopy already for the initial episode with resection of endometriosis lesions and closure of the diaphragmatic fenestrations. In case of repetition, one can either opt for a total thoracotomy with pleurectomy or hormonal therapy, or both together.

Hormonal treatment by Danatrol® (Danazol: anti-gonadotropin) which causes atrophy of endometrial cells is currently frequently used. Is prescribed at a dose of 600 to 800 mg daily for at least six months. However, several side effects observed as weight gain, virilization and mood disorders. The Decapeptyl® (similar GnRH: gonadotropin-releasing hormone analogue like) can also be used at 3.75 mg per month (i.m.) for six to nine months. These hormonal treatments can cause permanent sterility, which limits their use in women desiring pregnancy and promotes surgical management [19]. It is for this reason that your patient stopped medication.

Conclusion

It is important to make a good gynaecological history in all women with a pneumothorax, especially if the appearance is related to menstruation (24 to 48 hours before to 72 hours). If patients are known to endometriosis and pneumothorax is found on the right side, it is imperative to control the appearance of the diaphragm (tendon part) during any thoracoscopy.

If the diagnosis of catamenial pneumothorax is confirmed by pathology, it is wise to conduct ovarian hormone suppression. Among the different treatments, the pill is the one with the least side effects and should be offered as first-line, continuously taken, subject to its usual indications against. Progestin treatments are an alternative burdened with more side effects (bleeding, androgenic effects). If unsuccessful, a GnRH agonist therapy combined with a progestin or oestrogen plus progestin (add back therapy) to reduce bone loss and hypoestrogenic side effects may be offered on a 6 month period. Treatment on the longer term can be proposed, but requires a measurement of bone density annually.

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