



Catamenial Pneumothorax A case report and review of the literature.

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Summary

Catamenial pneumothorax is a spontaneous pneumothorax that occurs during menstruation. We present a case of catamenial pneumothorax which was treated with chemical pleurodesis after tube thoracostomy and underwater sealed drainage. The diagnosis was established based on the history. She currently does not have recurrence of pneumothorax after five months of follow up, but complains of monthly chest pain. The pathophysiology and the current research in the management of catamenial pneumothorax are reviewed.

Keywords : Catamenial pneumothorax-pleurodesis-endometriosis.

Introduction

Catamenial pneumothorax described by Maurer et al in 1958 and named by Lillington in 1972 is a recurrent syndrome of pneumothoraces that occurs between 48-72 hours after menstruation (1,7). It is a rare condition characterized by the recurring accumulation of air in the chest cavity (7). The condition occurs in the right lung in 90%-95% of cases (6,7). It never occurs during periods of no ovulation as in pregnancy or while on oral contraceptives and mainly affects women between 30-40 years (1). The diagnosis of this syndrome is difficult as it requires multiple pneumothoraces along with the recognition of a connection to the patient's menstrual cycle. A case of catamenial pneumothorax which was managed effectively with chemical pleurodesis and the current research in the management of this pathology are presented.

Case History

The patient is a 39year old female with an eight year history of intermittent chest pain at the back felt mainly at the right scapular region and sometimes radiating anteriorly beneath the right breast. The pain is piercing in nature and comes during her menstrual period. It is worse when she moves the chest and relieved by analgesics. The pain was associated with cough and on three occasions with breathlessness on exertion and had to be admitted.

She is nulliparous and undergoing treatment for primary infertility. She underwent myomectomy twice in 1996 and 1998 respectively.

On the three occasions that she presented at the hospital, she was not in respiratory distress. She was not pale and not cyanosed. Her respiratory rate was between 24-28 breaths per minute. The right chest was tympanic on percussion with diminished breath sounds.

Examination of the other systems was normal.

The chest X-ray on the third presentation is shown below in



Fig1. Right pneumothorax in patient with Catamenial pneumothorax

Tube thoracostomy with underwater-seal drainage was carried out during the three admissions. Chemical pleurodesis using 2 grams of tetracycline mixed with 50mls of 0.5% xylocaine was effected in her last two admissions. She still has the chest pain associated with cough every month but with no pneumothorax.

Discussion

Catamenial pneumothorax is defined as a recurrent syndrome of pneumothorax that occurs between 48-72hrs after menstruation (7). It mainly affects women between the ages of 30-40years with the history of pelvic endometriosis and the pneumothoraces are predominantly right-sided in 90-95% of cases (1,7). The diagnosis of this syndrome is difficult as it requires multiple pneumothoraces along with the recognition of a connection to the patient's menstrual cycle. As a result there have only been approximately 200 cases documented in the international literature as at April 2003 (7). Catamenial pneumothorax is a subset of primary spontaneous pneumothorax. The presentation is as with spontaneous pneumothorax i.e. chest pain and dyspnoea (5,7). In our patient in addition to the symptoms described she had cough which was recurrent with the menses.

The diagnosis of catamenial pneumothorax is usually based on either a chest pain associated with the start of menstruation or the history of recurring pneumothoraces that are predominantly right-sided and coincide with the onset of menstruation (5,7). The findings after invasive treatment usually support the diagnosis of catamenial pneumothorax. The diagnosis of our patient was mainly based on the history which is typical of catamenial pneumothorax (11). Four theories currently exist to explain the pathology of catamenial pneumothorax.

The most common is the movement of endometrial implants to the right diaphragm. This explanation is called the Anatomic model (7). The endometrial tissue preferentially goes to the right chest because of the recognized peritoneal circulation up from the pelvis to the right side (4,7). Other explanations include spikes of intraperitoneal pressure in the right subphrenic space accompanying respiratory activity and physical exertion with the solid liver acting as a piston. In contrast, the loose arrangement and the increased mobility of the left subphrenic organs minimized such pressure changes under the left hemi diaphragm (12,4).

These implants then create channels or fenestrations through the diaphragm that will allow further implants to move into the chest or allow transgression of air. These fenestrations are seen in only 19-38.8% of cases (2,7,14). The presumed timing of the pneumothorax around menstruation is postulated to occur because of the passage of the cervical mucous plug that allows retrograde movement of air. Studies have documented pneumoperitoneum at the same time as the

pneumothorax (4).

The second mechanism explaining the catamenial pneumothorax is known as the Metastatic Model. (7). In this model, the endometrial tissue makes its way from the pelvis to the chest cavity in one of two ways, either up through congenital diaphragmatic fenestrations or via micro embolism through the pelvic vein. Both modes of travel result in the movement of endometrial tissue into the thorax where it implants itself either onto the lung parenchyma and causes local defects or onto the diaphragm itself. (7). The third theory involves the elevated prostaglandin levels that occur during menstruation. Of significant interest is prostacycline F2, which causes severe vasospasm and bronchospasm. It is believed that this leads to the rupture of alveoli and results in a pneumothorax (7). The last theory is the spontaneous rupture of sub pleural blebs due to hormone changes occurring with menses that leads to pneumothorax. These last two theories do not explain the right sided predominance of catamenial pneumothorax. One expects to find burst or unburst blebs on the lung surfaces but this is not seen on thoracoscopic investigations considering the last theory.

In most cases of catamenial pneumothorax, the initial pneumothorax episodes are treated in the same way as any primary pneumothorax. When this fails medication and hormonal drugs (e.g. Danazol, gonadotrophin-releasing hormone (GnRH) analogues, possibly hormonal contraceptive drugs or progestogens.) with effective pleurodesis are considered to be the most efficient methods of treatment.(3,6,9).With our case the use of hormonal drugs was not advisable since she is being treated for infertility.

Over the years, two main categories of treatment have been tried - medical and surgical. The medical options are mainly hormonal therapies that are aimed at suppressing the activity and growth of endometrial tissue present in the chest. This is done by suppressing ovulation and oestrogen release (7,10). The hormonal treatments that have been tried to date are testosterone derivatives, oral contraceptives and GnRH agonists. They have been found to prevent approximately 50% of recurrences of pneumothorax (7). GnRH agonists are known to preserve the reproductive potential of the patient (7,8).

Surgical options include pleurodesis, hysterectomy, and tubal ligation, thoracoscopic diaphragmatic endometrial resection along with bleb and apical resection where applicable (3,5,7). Pleurodesis (mechanical or chemical) have been found to be an effective method of preventing recurrent pneumothoraces but does not treat the root problem. In some centers, pleurodesis has been used as a first line of treatment for primary spontaneous pneumothorax. For patients with catamenial pneumothorax, it is common for them to still experience chest pain and other symptoms without lung collapse during menses after pleurodesis. (4,7). Our patient still has

chest pain with cough during menses but without pneumothorax. This is known to occur when there is a remaining endometrial tissue in the pleural cavity.(7) Hysterectomy with bilateral oophorectomy is another option for treatment that is most often used in women outside of childbearing years. It is effective for sometime but has the drawback of hypoestrogenia. On the bases of the hypothesis of transfallopian passage of air, some patients are treated by tubal ligation only (3,5). This mode of treatment is not suitable for patient s in their child bearing years.

Evaluation of the diaphragm and the pleural space are easily done with the thoracoscope and if holes or endometrial implants are seen, they are managed by resection and/or suturing (4,7). Multiple holes over large areas of the diaphragm are best suited for pleurodesis. Localized areas may be resected or sutured. Large resection may be patched with a prosthetic material (4). Plication of the involved area has also been documented (14). If endometrial implants are found their resection should be performed to limit further endometrial spreading. It has been found that overall the surgical treatment of catamenial pneumothorax results in a far lower recurrence rate than the hormonal treatment does (7). Surgical treatment should be accomplished during menstruation for an optimal visualization of pleurodiaphragmatic endometriosis.

Conclusion

To our knowledge this is the first case of catamenial pneumothorax reported in Ghana. The diagnosis is mainly based on the history and clinical findings. The case discussed has the typical history and clinical findings of catamenial pneumothorax. Management of catamenial pneumothorax is either surgical or hormonal or the combination of both. The combination therapy is the most efficient way of managing this condition. It is more difficult with patients in their child bearing years since hormonal therapy becomes unsuitable. Video-assisted thoracoscopic surgery is helpful if available.

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