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Catamenial Pneumothorax: Menstruation-Associated Pneumothorax

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ABSTRACT

Introduction: This article explores catamenial pneumothorax, a rare form of spontaneous pneumothorax linked to the menstrual cycle and most commonly associated with thoracic endometriosis syndrome. It discusses the current understanding of its epidemiology, pathophysiology, clinical presentation, diagnostic challenges, and treatment strategies. The discussion underscores the importance of accurate diagnosis and multidisciplinary treatment approaches to reduce recurrence and improve quality of life.

Materials and Methods: A thorough literature search was performed using PubMed and Google Scholar databases with the following keywords: “catamenial pneumothorax”, “endometriosis”, “VATS”, “hormone therapy”.

Summary: Catamenial pneumothorax (CP) is a rare, cyclical spontaneous pneumothorax occurring predominantly in reproductive-age women, often associated with thoracic endometriosis syndrome. Its exact pathophysiology remains unclear, but proposed mechanisms include metastatic spread of endometrial tissue, hormonal influences, and anatomical defects in the diaphragm, especially on the right side. Diagnosis relies on clinical suspicion, imaging, and video-assisted thoracoscopic surgery, while effective management typically involves surgical intervention combined with postoperative hormonal therapy to reduce recurrence.

Conclusions: Catamenial pneumothorax is a significant clinical manifestation of thoracic endometriosis and should be considered in reproductive-age women presenting with recurrent pneumothorax correlated with their menstrual cycle. Optimal outcomes are achieved through a combination of video-assisted thoracoscopic surgery and postoperative hormonal therapy, supported by a multidisciplinary approach.

Keywords: catamenial pneumothorax, endometriosis, VATS, hormone therapy.

Introduction

Catamenial pneumothorax (CP) is a rare clinical entity most often linked to thoracic endometriosis syndrome (TES), which involves the presence of endometrial tissue in the pleura and lung parenchyma. The term “catamenial pneumothorax” was first described by Maurer in 1958 and later coined by Lillington in 1972 [1]. CP is defined by its cyclical recurrence,

typically occurring from 24 hours prior to up to 72 hours following the onset of menstruation. It typically presents as a recurring spontaneous pneumothorax in women of reproductive age, predominantly involving the right pleural cavity (92%) and is usually associated with the menstrual cycle, though not necessarily with every cycle [2].

Although a correlation with thoracic endometriosis has been established, the precise pathophysiological mechanisms remain poorly elucidated [3]. CP is believed to primarily affect ovulating women and is generally not seen in those who are pregnant, experiencing menarche, or using ovulation-suppressing treatments. Nevertheless, rare cases have been reported in women undergoing ovulatory suppression and even during pregnancy [4]. The condition is the most frequent manifestation of thoracic endometriosis syndrome, which also includes four other cyclical clinical forms: catamenial hemothorax, catamenial hemoptysis, pulmonary endometriotic nodules, and catamenial chest pain [5]. The diagnosis requires a high index of clinical suspicion, particularly in women of reproductive age presenting with cyclical chest pain or dyspnea. A thorough gynecological history is essential [6]. While imaging modalities can assist in diagnosis, video-assisted thoracoscopic surgery (VATS) remains the gold standard for both diagnosis and treatment. Surgical management combined with a minimum of six months of hormonal therapy has been shown to improve outcomes and reduce recurrence rates [1]. Treatment also focuses on managing associated pelvic endometriosis, if present, using hormonal suppression [7]. Although VATS is effective, recurrence has been reported in approximately 25% of patients despite appropriate treatment [8].

Epidemiology

Catamenial pneumothorax accounts for 30–73% of thoracic endometriosis syndrome cases and may represent 18–33% of spontaneous pneumothorax cases in women [2,9]. The condition is considered rare [6]. Women diagnosed with CP typically fall within the age range of 34 to 37 years, which differs from the general demographic for primary spontaneous pneumothorax [10]. Endometriosis itself affects approximately 3–10% of women of reproductive age and 2–5% of postmenopausal women, but thoracic endometriosis and CP remain underdiagnosed and often experience delayed identification [9]. In some studies, patients have been categorized based on the presence or absence of diaphragmatic defects, with such defects observed in up to 50% of cases [11].\

Endometriosis

Catamenial pneumothorax is linked to thoracic and pelvic endometriosis, though not always confirmed. Endometriosis involves endometrial tissue (glands and stroma) growing outside the uterus. Its cause is unclear, and treatment can be challenging. The pelvic form is most common, typically causing pelvic pain and infertility. Thoracic endometriosis, the most frequent site outside the pelvis, occurs when this tissue is found in the chest. It is confirmed by finding both glands and stroma, or considered probable if only stroma is present [12]. Catamenial pneumothorax represents a clinical manifestation of thoracic endometriosis syndrome; however, histopathological confirmation of endometrial deposits within the thoracic cavity - defining thoracic endometriosis-related catamenial pneumothorax - has been reported in 52-87% of surgically treated cases. Consequently, the diagnosis of catamenial pneumothorax is predominantly established on clinical criteria, whereas a definitive diagnosis of thoracic endometriosis-related catamenial pneumothorax necessitates meticulous intraoperative evaluation [2].

Etiology

The link between spontaneous pneumothorax and the menstrual cycle is well known, but its exact cause remains unclear. Three main theories explain this: metastatic, hormonal, and anatomical. The metastatic theory suggests that endometrial tissue spreads to the pleural space through lymphatic channels, diaphragmatic defects, or blood, mostly affecting the right side due to congenital diaphragmatic openings. This tissue causes irritation and air leaks, leading to pneumothorax, supported by findings of endometrial deposits in 13% to 62.5% of cases. The hormonal theory proposes that high prostaglandin F2 levels during ovulation cause lung vasospasm and bronchospasm, resulting in alveolar rupture and pneumothorax. However, this theory lacks support as anti-inflammatory treatments fail to prevent recurrence and it doesn’t explain the right-sided dominance. The anatomical theory suggests air enters the pleural space from the peritoneal cavity through diaphragmatic fenestrations, triggered by the loss of the cervical mucus plug during menstruation. This theory is supported by radiographic evidence, the high incidence of diaphragmatic defects in patients, and successful prevention of recurrence after surgical repair of these defects, particularly on the right side [13]. Catamenial pneumothorax mainly affects the right side because the clockwise movement of peritoneal fluid

from the pelvis to the right subphrenic area carries endometrial implants to the right diaphragm. The liver's 'piston effect' then passes sudden increases in abdominal pressure through existing holes in the diaphragm, pushing air into the pleural cavity [14].

Symptoms

Catamenial pneumothorax typically presents with symptoms resembling a pneumothorax that appear in connection with menstruation, usually starting about 24 hours before and continuing up to 48–72 hours after its onset. However, these symptoms might not be present during every menstrual cycle [9]. The symptoms they experience are similar to those of spontaneous pneumothorax and include pleuritic chest pain, coughing, and shortness of breath [10]. Pain in the scapular or thoracic region during menstruation often occurs before the onset of pneumothorax [15]. Irritation of the diaphragm can cause referred pain to the area around the shoulder blade or radiate to the neck, usually on the right side. In most cases (95%), the right side of the chest is affected, while 5% involve the left side, and 3% show bilateral involvement [10]. A background of infertility, ongoing pelvic discomfort, and symptoms like painful menstruation or intercourse could indicate thoracic endometriosis syndrome (TES). Occasionally, this condition may also involve a diaphragmatic hernia linked to endometriosis [9]. A history of coughing up blood during menstruation can be considered an indicator of pulmonary endometriosis [16]. Catamenial pneumothorax are generally mild to moderate in severity, though in rare instances, such as with extensive thoracic endometriosis following prior surgeries, they can become life-threatening [12].

Radiological diagnostics

Currently, no definitive diagnostic criteria exist for this condition. Radiologically, catamenial pneumothorax may be suspected in the presence of pneumoperitoneum accompanied by right-sided pneumothorax. Additional diagnostic indicators include extensive diaphragmatic defects, diaphragmatic hernias involving hepatic tissue, and small diaphragmatic perforations characterized as "air-filled bubbles" corresponding to diaphragmatic fenestrations. Computed tomography has the capability to identify endometrial nodules located on the diaphragmatic surface. Although magnetic resonance imaging may enhance the detection of endometriotic

lesions, its routine application in the diagnostic evaluation of pneumothorax cases is currently not substantiated [17].

Management

The treatment consists of two primary approaches: conservative management, which involves hormone replacement therapy to reduce the risk of pneumothorax and hemothorax recurrence; and surgical intervention, recommended when hormone therapy is ineffective, causes significant side effects, or when symptoms return after stopping treatment [18]. Management of catamenial pneumothorax primarily involves surgical intervention, preferably via video-assisted thoracoscopic surgery (VATS), to facilitate histopathological confirmation of thoracic endometriosis and to address the underlying pathophysiological mechanisms contributing to pneumothorax [19]. In cases of catamenial pneumothorax, it is likely even more beneficial, as it enables thorough examination of the chest cavity, including the diaphragm, which is challenging to inspect through a restricted axillary thoracotomy [20].

Standard surgical procedures often include partial diaphragmatic resection, excision of endometriotic lesions from the visceral pleura, and pleurodesis with talc. Postoperative hormonal therapy aimed at inducing ovarian suppression is imperative. A multidisciplinary, multimodal therapeutic strategy remains essential for optimizing clinical outcomes in this complex condition [19]. Hormonal therapy used alongside surgery helps to prevent the recurrence of catamenial and/or endometriosis-associated pneumothorax. It is recommended that all patients diagnosed with catamenial and/or endometriosis-related pneumothorax receive multidisciplinary care and treatment with a gonadotrophin-releasing hormone (GnRH) analogue, which induces amenorrhea, for 6 to 12 months immediately following surgery [17]. Medical treatment alone does not appear to be effective, with recurrent CP rates as high as 50%. Concomitant surgery and hormone therapy significantly reduces this risk [21].

Summary

Catamenial pneumothorax (CP) is a rare but clinically significant condition, most often representing the thoracic manifestation of endometriosis. It predominantly affects women of reproductive age and is characterized by the cyclical onset of spontaneous pneumothorax in close temporal relation to menstruation, typically involving the right pleural cavity. Despite its increasing recognition, CP remains underdiagnosed due to the nonspecific nature of its

symptoms and the lack of standardized diagnostic criteria. A high index of suspicion, especially in women with recurrent pneumothorax and pelvic endometriosis symptoms, is critical for timely diagnosis. The etiology of CP is multifactorial and still not fully understood, with leading theories including metastatic spread of endometrial tissue, hormonal influences, and anatomical pathways allowing air to migrate from the abdominal to the pleural cavity. The presence of diaphragmatic fenestrations and endometrial implants in the thoracic cavity supports the anatomical and metastatic hypotheses. Radiological findings can aid in the diagnostic process, but video-assisted thoracoscopic surgery (VATS) remains the gold standard for both diagnosis and treatment, enabling direct visualization and removal of ectopic endometrial tissue, repair of diaphragmatic defects, and tissue sampling for histopathology. Management of CP is best approached through a multidisciplinary strategy that combines surgical intervention with postoperative hormonal therapy to suppress ovulation and reduce the risk of recurrence. Hormonal suppression, most often with GnRH analogues, is recommended for at least 6 to 12 months following surgery. Isolated hormonal therapy without surgery has limited success and carries a high recurrence rate.

In conclusion, catamenial pneumothorax, though rare, should be considered in the differential diagnosis of recurrent pneumothorax in menstruating women, particularly when right-sided and temporally associated with menses. Early recognition, surgical intervention via VATS, and long-term hormonal suppression constitute the cornerstone of effective management. Improved awareness among clinicians and gynecologists, along with collaborative care, is essential to reduce diagnostic delays and optimize outcomes for affected patients.

Disclosure

Author's contribution

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