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Case Report

Catamenial pneumothorax: a rare manifestation of endometriosis[☆]

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ABSTRACT

Endometriosis is a common gynecological disease that primarily affects premenopausal women. It is mainly found in the pelvis but may be found at several extrapelvic locations. Thoracic endometriosis is a rare extrapelvic location of endometriosis and the leading cause of catamenial pneumothorax. We describe the case of a 35-year-old woman with a background of pelvic pain presenting to the emergency department with chest pain and dyspnea. The chest X-ray in the emergency department showed a large right-sided pneumothorax. Further imaging studies during patient evaluation revealed extensive fibrotic changes in the pelvis and well-defined solid nodules with high signal on T2 and T1-weighted images on MRI in abdominal and thoracic locations, rendering the diagnosis of a catamenial pneumothorax in a patient with pelvic, abdominal and thoracic endometriosis.

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Introduction

Endometriosis is a common gynecological disease that predominantly affects women of reproductive age. It is defined by the presence of functional ectopic endometrial tissue and stroma outside the uterus. The ectopic endometrial tissue responds to hormonal stimulation with various degrees of cyclic bleeding, inducing an inflammatory response that may progress to fibrosis [1,2].

Endometriosis is primarily found in the pelvis. Less commonly, endometriosis may be found at several extrapelvic locations. Thoracic endometriosis is an exceedingly rare loca-

tion for endometriosis and the leading cause of catamenial pneumothorax [3].

Herein we describe the case of a 35-year-old nulliparous woman with a background of repeated episodes of pneumothorax and long-term pelvic pain of unknown origin. Radiologic studies revealed a large right-sided pneumothorax, extensive fibrotic changes in the pelvis, and well-defined solid nodules with high signal on T2 and T1-weighted images on MRI in abdominal and thoracic locations.

A diagnosis of pelvic, abdominal, and thoracic endometriosis complicated by repeated catamenial pneumothoraxes was proposed.

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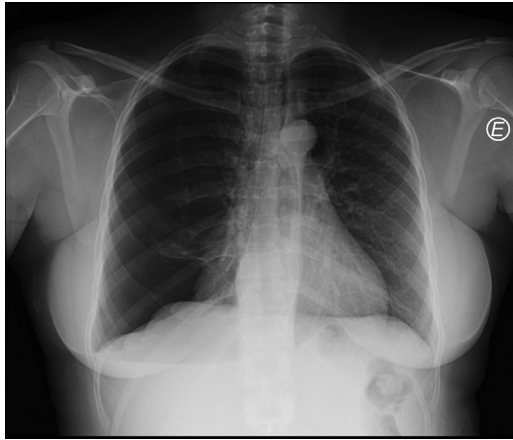


Fig. 1 – Chest X-ray obtain at admission to the emergency department shows a large volume right-sided pneumothorax.

Case report

A 35-year-old premenopausal nulliparous woman presented with chest pain and dyspnea to the emergency department. She refers to several similar, although less severe, episodes in the past months. Like the current episode, these episodes coincide with the first days of menstruation.

Clinical examination revealed tachypnea and decreased breath sounds over the right hemithorax. Laboratory results were unremarkable.

When asked about her medical background, the patient complains about long-term pelvic pain, with no additional relevant personal or familial medical histories.

A chest X-ray showed a large volume right-sided pneumothorax (Fig. 1). The chest X-ray also revealed increased right intercostal spaces compared to the left hemithorax and a slight depression of the right hemidiaphragm, suggesting a potential tension pneumothorax, leading to tube drainage in the emergency department.

After tube drainage, a chest CT was performed, showing 2 well-defined solid nodules in the right cardiophrenic angle and in the hepatorenal space (Figs. 2 and 3).

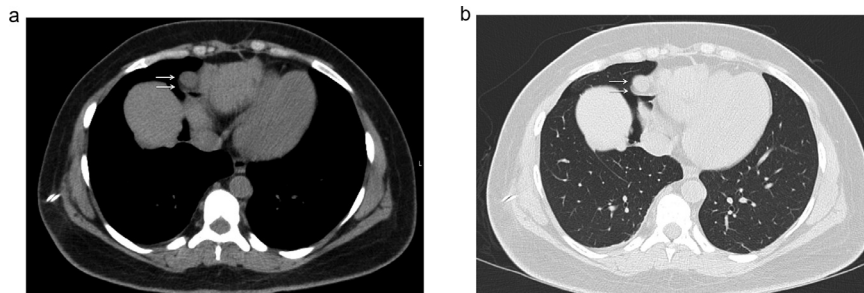


Fig. 2 – Chest CT (mediastinum W [a], lungs W [b]) shows a round, well-demarcated nodule in the right cardiophrenic angle (arrows).

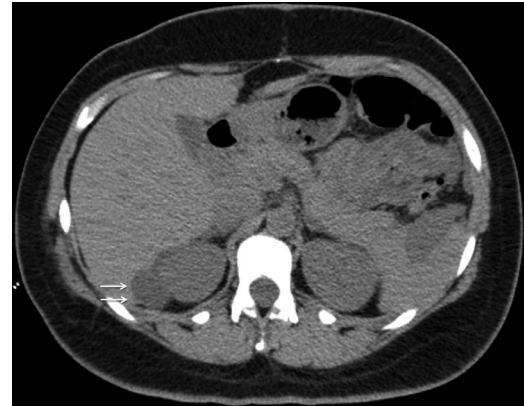


Fig. 3 – A well-demarcated solid nodule is found in the right hepatorenal space in the upper abdominal segments intercepted in the chest CT study (arrows).

A transvaginal ultrasound (US) and a pelvic MRI were performed to clarify the background of chronic pelvic pain. Transvaginal US depicted an enlarged, non-mobile, uterus with several leiomyomas. Two ill-defined, hypoechoic nodular masses were found in the vesicouterine pouch and in the rectouterine pouch. A fluid-filled, dilated fallopian tube was found in the right adnexal region. MRI depicted extensive fibrotic changes in the pelvis, involving all three compartments with endometriotic plaques located in the vesicouterine pouch adjacent to the bladder dome (Fig. 4) and in the rectouterine pouch (Fig. 5), associated with a dilated blood-filled right fallopian tube (Fig. 6), loculated hematic ascites (Fig. 7), and signs of "frozen pelvis". Several uterine leiomyomas were found.

Chest MRI to clarify the findings depicted in chest CT showed multiple pleural-based millimetric solid nodules with high signal both on T2- and T1-weighted images. Larger nodules with matching features were found in the right cardiophrenic angle and in the right hemidiaphragm (Figs. 8–10). A similar nodule was depicted in the right hepatorenal space (Fig. 11).

Clinical and radiologic findings rendered the diagnosis of pelvic, abdominal, and thoracic endometriosis with associated catamenial pneumothorax.

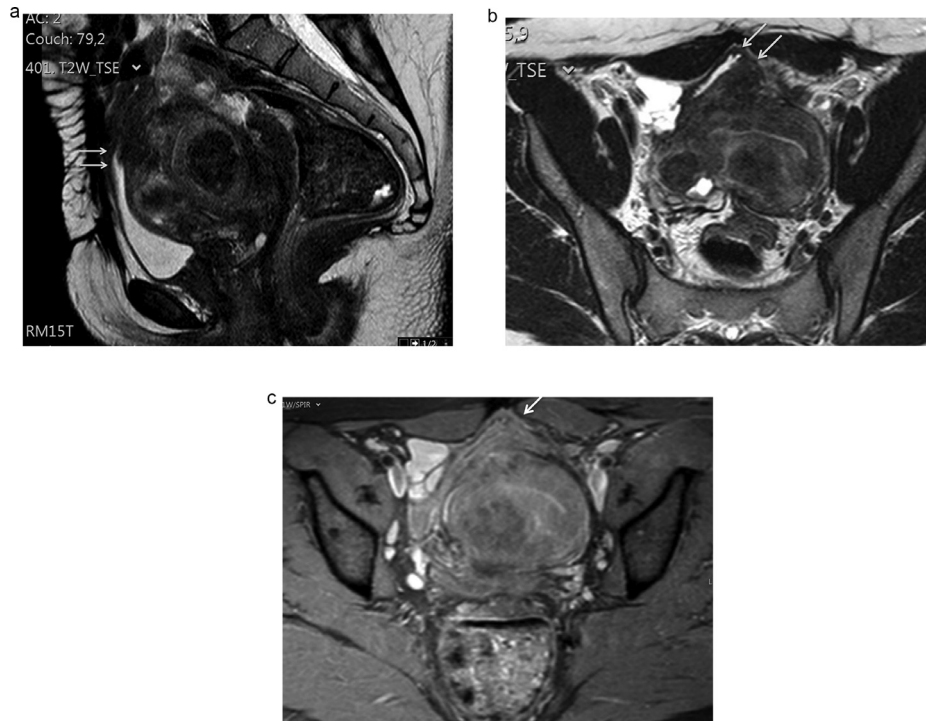


Fig. 4 – Sagittal T2-weighted MRI (a) demonstrates endometriosis involving the anterior compartment of the pelvis, infiltrating the superior portion of the bladder (arrows). Axial T2-weighted MRI (b) and axial T1-weighted MRI with fat saturation (c) show encasement of the superior portion of the bladder (arrows).

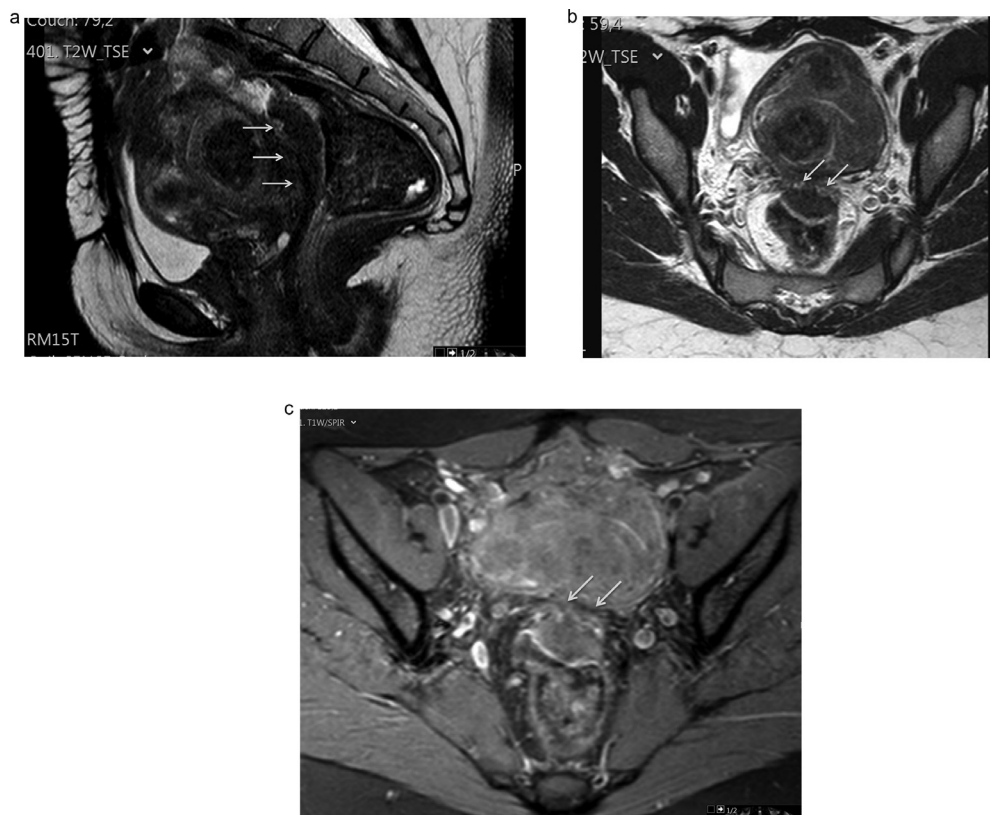


Fig. 5 – Sagittal T2-weighted MRI (a), axial T2-weighted MRI (b), and T1-weighted MRI with fat saturation (c) show involvement of the posterior compartment of the pelvis with endometriosis in the posterior *cul-de-sac* with infiltration of the rectal wall.

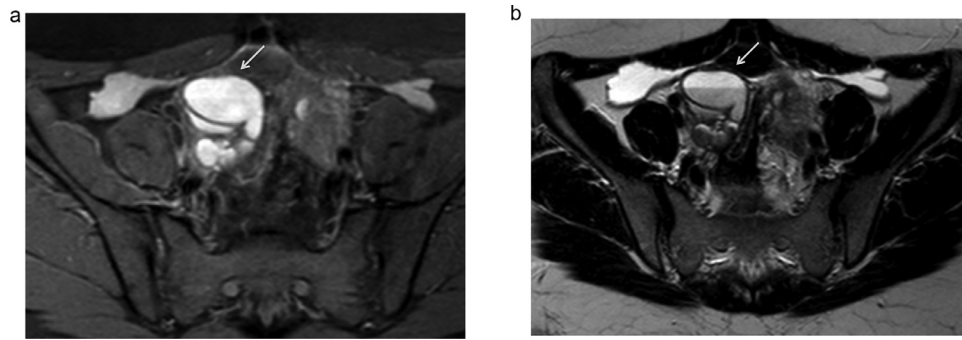


Fig. 6 – Pelvic MRI shows a dilated right fallopian tube (arrows). The distended tube has high signal intensity on T1-weighted fat-suppressed imaging due to the presence of blood products (a). A fluid-fluid level can be seen in T2-weighted imaging (b).

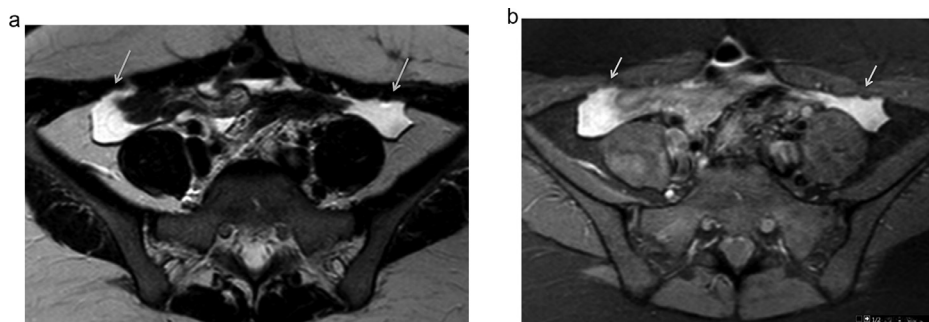


Fig. 7 – Pelvic MRI axial T2-weighted (a) MRI shows loculated ascites. The fluid has high signal intensity on T1-weighted fat-suppressed imaging due to the presence of blood products (b).

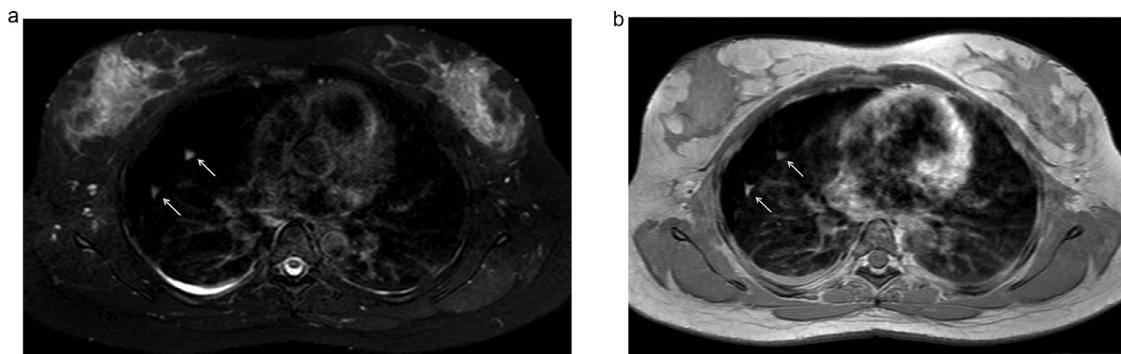


Fig. 8 – Chest MRI shows several millimetric right pleural-based nodules (arrows). The nodules have high signal intensity both on T2 (a) and T1-weighted imaging (b).

Discussion

Endometriosis is an estrogen-dependent benign gynecologic disease defined by the presence of functional ectopic endometrial tissue and stroma outside the uterus, which primarily affects premenopausal women and is associated with signif-

icant morbidity. The ectopic endometrial tissue responds to hormonal stimulation with various degrees of cyclic bleeding, which induces an inflammatory response that may progress to fibrosis [1,2].

Endometriosis is mainly found in the pelvis. It can involve the anterior, middle, and posterior pelvic compartments, affecting the ovaries, fallopian tubes, uterus, uterosacral liga-

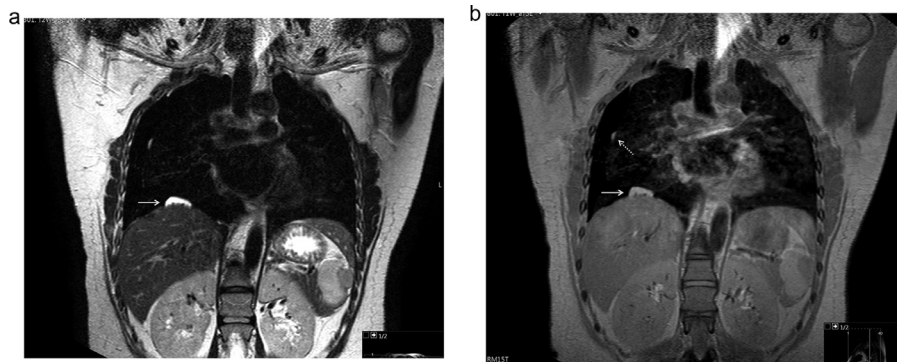


Fig. 9 – Coronal T2-weighted MRI (a) and T1-weighted MRI (b) show a large diaphragmatic nodule with high signal intensity on both sequences (arrows). On T1-weighted imaging, a high signal pleural-based millimetric nodule is noticed (dashed arrow).

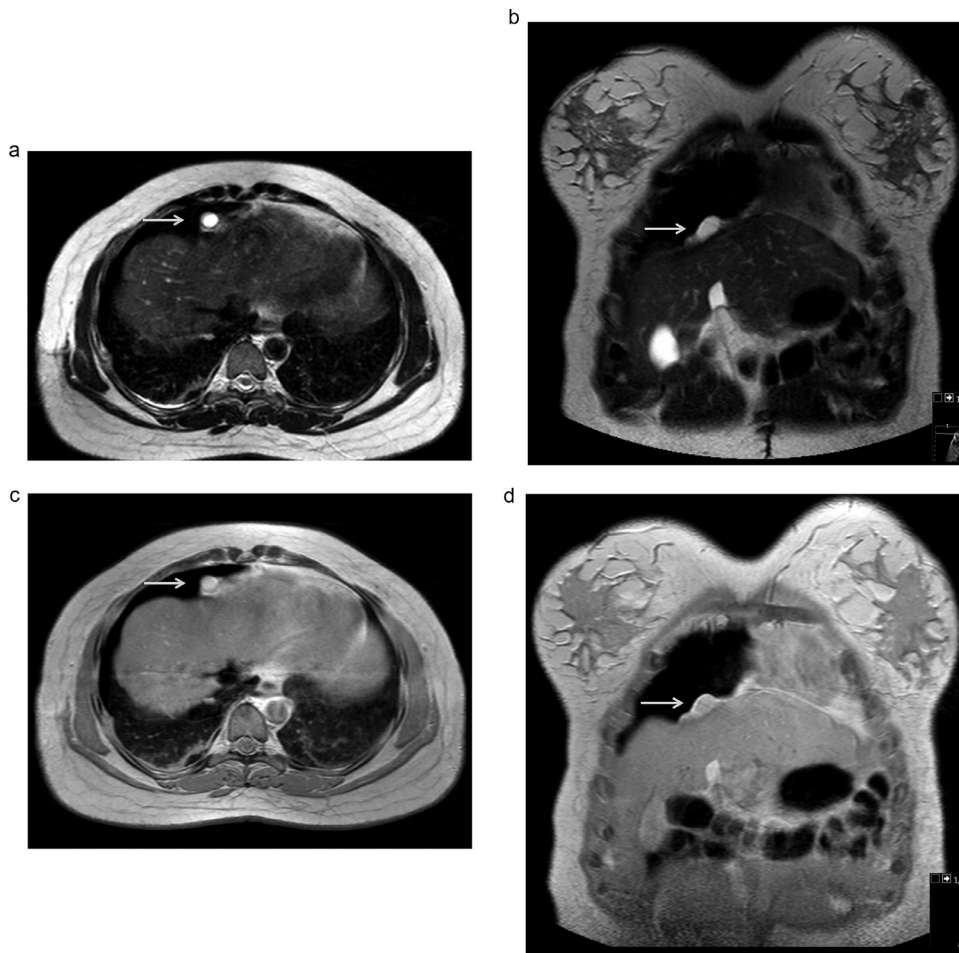


Fig. 10 – Axial and coronal T2-weighted imaging (a, b) and T1-weighted imaging (c, d) show a large nodule with high signal intensity on both sequences in the right cardiophrenic angle.

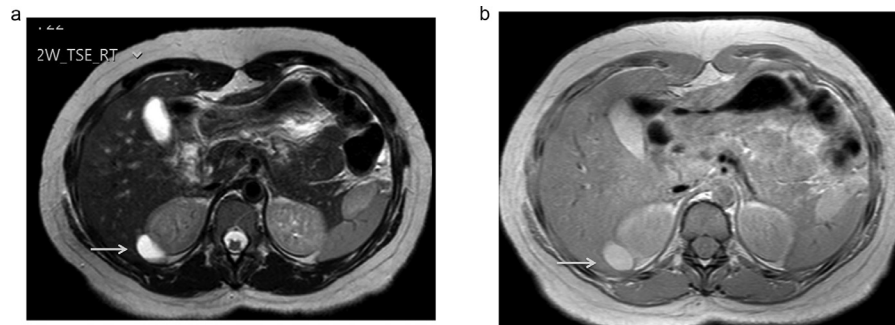


Fig. 11 – A well-demarcated solid nodule is found in the right hepatorenal space (arrows) in the upper abdominal segments intercepted in the chest MRI study. The nodule has a high signal both in T2 (a) and T1-weighted imaging (b).

ments, round ligaments, broad ligaments, *cul-de-sac*, ovarian fossa, appendix, large bowel, bladder, ureters, and rectovaginal septum [4].

Extra pelvic locations can be involved. Extrapelvic endometriosis may be found at several sites, including the abdominal wall, vulva and perineum, liver, skin, central nervous system, and the thorax. Thoracic endometriosis, defined as the presence of ectopic endometrial tissue inside the thoracic cavity, can affect the pleura, lung parenchyma, airways, and diaphragm [3]. It is an exceedingly rare location for endometriosis and the leading cause of catamenial pneumothorax.

Clinical manifestations depend on the anatomic locations of endometriosis: pelvic endometriosis causes pelvic pain and infertility; thoracic endometriosis can manifest as catamenial pleuritic chest pain, hemoptysis, hemothorax, or pneumothorax. Pneumothorax is the most common manifestation of thoracic endometriosis (80% of cases) [5].

Catamenial pneumothorax is defined as recurrent pneumothorax associated with menstruation. The right side is most often involved (85%-95%) [3], but catamenial pneumothorax can be left-sided or bilateral [3]. Although by definition, catamenial pneumothorax occurs in the menstrual period, it does not happen with every menstrual cycle [5]. Thoracic endometriosis can occur in isolation, but it is usually associated with extensive endometriosis involving the reproductive, genitourinary, and gastrointestinal systems [6].

Although the combination of laparoscopy and histological verification of endometrial glands and stroma is considered the gold standard for diagnosing endometriosis, [7] radiologic studies are paramount in supporting the diagnosis of endometriosis, staging the disease, assisting with surgical planning, and excluding other conditions.

US and MRI are the most frequently used imaging methods [8]. Transvaginal-US is usually the first-line imaging modality in assessing pelvic endometriosis. However, US findings are variable and may mimic those of other benign and malignant gynecologic conditions.

MRI has a high sensitivity for detecting blood products and can identify sites of disease hidden by dense adhesions.

In the pelvis, a dilated fallopian tube retaining hemorrhagic content - hematosalpinx - is often associated with endometriosis [9]. Deep endometriosis lesions usually have ill-defined margins and low T2 signal due to fibrosis. Subcentime-

ter foci with high T2 signal depict ectopic endometrial glands within these infiltrating fibrotic masses [9].

In the thorax, the MRI appearance of pleural-based lesions exhibiting homogeneous high signal both on T2- and T1-weighted images is characteristic of pleural endometriosis [10].

Both medical and surgical approaches can be used to manage thoracic endometriosis patients. Medical therapy suppresses the active ectopic endometrial tissue using progestins, danazol, or gonadotropin-releasing hormone analogs [11]. Surgical approaches are employed when medical treatment fails. Video-assisted thoracoscopic surgery or open surgery is performed to clear the ectopic endometrial tissue [11]. Chemical pleurodesis can help manage catamenial pneumothorax in such cases [11].

Conclusion

Endometriosis is a common gynecological disease mainly found in the pelvis causing pelvic pain and infertility. Less frequently, extra pelvic locations, such as the thorax, can be involved.

Catamenial pneumothorax in the setting of thoracic endometriosis should be considered in premenopausal women with recurrent right-sided chest pain associated with menstruation, especially when a history of concurrent pelvic endometriosis is present.

Thoracic MRI showing pleural-based lesions exhibiting homogeneous high signal on T2- and T1-weighted images is highly suggestive of thoracic endometriosis and should be performed in premenopausal women with catamenial pleuritic chest pain, hemoptysis, hemothorax, or pneumothorax.

Patient consent statement

The patient's informed consent for the publication of this case was granted.

There are no ethical issues for the publication of this case report according to the standard of our institution.

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