

Delay in diagnosis of endometriosis:

a case report of catamenial pneumothorax

INTRODUCTION

Endometriosis is a common gynaecological condition where endometrium-like tissue is located outside the uterine cavity, predominantly inside the pelvis, causing symptoms such as dysmenorrhoea and dyspareunia.¹ However, extrapelvic endometriosis can also occur, often leading to delay in diagnosis that results in chronic symptoms and progression of the disease with possible lifelong consequences (for example, infertility).¹⁻³ The current case report illustrates a rare example of extrapelvic endometriosis and catamenial pneumothorax (that is, pneumothorax occurring during the perimenstrual period). The aim of this article is therefore to improve endometriosis awareness among GPs. The key message from this case report is that GPs should be aware of endometriosis when women present with symptoms that fluctuate with their menstrual cycle.

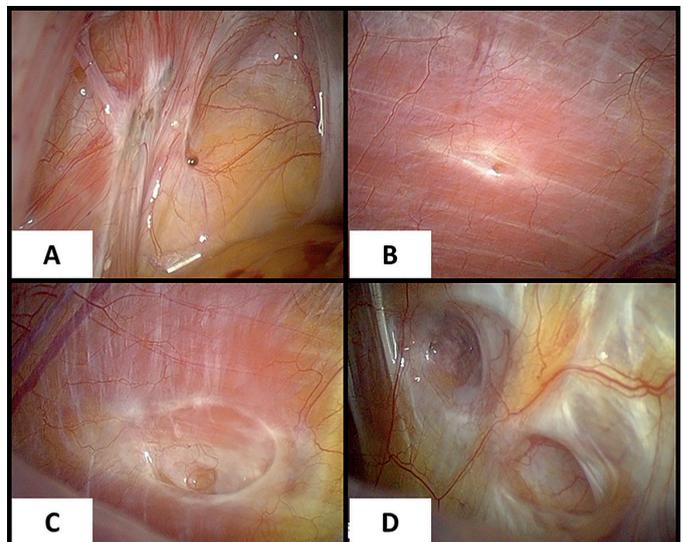
CASE REPORT

A 34-year-old woman was referred by her GP because of coughing, painful breathing, and dyspnoea. A right-sided pneumothorax was diagnosed and successfully drained. After 5 months, a second pneumothorax occurred, for which a thoracoscopic talc pleurodesis was performed to obliterate the pleural space to prevent further recurrences. No abnormalities were seen during thoracoscopy or chest CT scan. During the follow-up visit at the

pulmonologist, the patient indicated that she had noticed that the thoracic pain always started after the beginning of her period. Moreover, retrospectively, the series of pneumothoraxes started when the patient switched from an oral contraceptive to a levonorgestrel intrauterine device. A catamenial pneumothorax was considered and the patient was referred to the gynaecologist. In the past, an oophorectomy had been performed due to recurrent ovarian cysts, but no endometriosis was noted. Now, she reported minor cyclic abdominal pain, without dyspareunia, dysmenorrhoea, or bladder or bowel dysfunction, and physical examination revealed no signs of endometriosis. Despite ovarian suppression by a continuous progestin, the patient developed another pneumothorax and a video-assisted thoracoscopic surgery (VATS) was performed with a partial pleurectomy. No abnormalities were seen during the procedure; moreover, no indications of endometriosis were spotted (for example, bullae or fenestrations in the diaphragm). Pathological examination of

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Figure 1. Endometriosis with extensive scarring in the pelvis (A); endometriosis spots (B); fenestration (C); and scarring on the diaphragm (D).



Box 1. Possible location of endometrioses for women presenting with cyclic symptoms

Gynaecological tract	Menarche, abnormal bleeding, infertility, pregnancies, dysmenorrhoea, dyspareunia, pelvic pain
Pulmonary tract	Chest pain, pneumothorax, haemothorax, haemoptysis
Gastrointestinal tract	Dyschezia, constipation, diarrhoea, rectal bleeding
Urinary tract	Frequency, urgency, haematuria, dysuria

REFERENCES

1. Giudice LC, Kao LC. Endometriosis. *Lancet* 2004; **364**(9447): 1789–1799.
2. Burton C, Iversen L, Bhattacharya S, *et al*. Pointers to earlier diagnosis of endometriosis: a nested case-control study using primary care electronic health records. *Br J Gen Pract* 2017; DOI: <https://doi.org/10.3399/bjgp17X693497>.
3. Machairiotis N, Stylianaki A, Dryllis G, *et al*. Extrapelvic endometriosis: a rare entity or an under diagnosed condition? *Diagn Pathol* 2013; **8**: 194.
4. Davis AC, Goldberg JM. Extrapelvic endometriosis. *Semin Reprod Med* 2017; **35**(1): 98–101.
5. Dunselman GA, Vermeulen N, Becker C, *et al*. ESHRE guideline: management of women with endometriosis. *Hum Reprod* 2014; **29**(3): 400–412.
6. Gabriel B, Nassif J, Trompoukis P, *et al*. Prevalence and management of urinary tract endometriosis: a clinical case series. *Urology* 2011; **78**(6): 1269–1274. <http://dx.doi.org/10.1016/j.urology.2011.07.1403>.
7. Leone Roberti Maggiore U, Ferrero S, Candiani M, *et al*. Bladder endometriosis: a systematic review of pathogenesis, diagnosis, treatment, impact on fertility, and risk of malignant transformation. *Eur Urol* 2017; **71**(5): 790–807.
8. Nezhat C, Li A, Falik R, *et al*. Bowel endometriosis diagnosis and management. *Am J Obstet Gynecol* 2018; **218**(6): 549–562. <https://doi.org/10.1016/j.ajog.2017.09.023>.
9. Marshall MB, Ahmed Z, Kucharczuk JC, *et al*. Catamenial pneumothorax: optimal hormonal and surgical management. *Eur J Cardiothorac Surg* 2005; **27**(4): 662–666.
10. Lillington GA, Mitchell SP, Wood GA. Catamenial pneumothorax. *JAMA* 1972; **219**(10): 1328–1332.
11. Andolf E, Thorsell M, Källén K. Caesarean section and risk for endometriosis: a prospective cohort study of Swedish registries. *BJOG* 2013; **120**(9): 1061–1065.
12. Johnston JL, Reid H, Hunter D. Diagnosing endometriosis in primary care: clinical update. *Br J Gen Pract* 2015; DOI: <https://doi.org/10.3399/bjgp15X683665>.
13. Pugsley Z, Ballard K. Management of endometriosis in general practice: the pathway to diagnosis. *Br J Gen Pract* 2007; **57**(539): 470–476.
14. Bulun SE. Endometriosis. *N Engl J Med* 2009; **360**(3): 268–279.

the pleural tubes demonstrated no signs of endometriosis. After the VATS, the patients suffered from three more recurrences of a pneumothorax. After the sixth right-sided pneumothorax, a laparoscopy was performed to search for endometriosis and/or fenestrations in the diaphragm. During laparoscopy, peritoneal endometriosis and endometriotic lesions with extensive scarring and fenestrations were seen at the diaphragm (Figure 1). Three years later, a partial pleurectomy with pleurodesis (that is, obliteration of the pleural space) was finally performed because continuous hormonal suppression was unsuccessful. At present, there has been no recurrence of a pneumothorax; nevertheless, once every few months the patient still has a pulling and painful sensation in her thorax.

DISCUSSION

Endometriosis can affect various organ systems and symptoms depending on the location, resulting in complaints of the gynaecological, urinary, gastrointestinal, or pulmonary tract (Box 1).^{3,4} A common misconception among GPs is that endometriosis is rare in young women; however, endometriosis can also occur in teenagers and adolescents.⁵ Principal symptoms include dysmenorrhoea, chronic (menstrual or continuous) pelvic pain, dyspareunia, and abnormal bleeding.¹ Endometriosis located in the urinary tract can lead to symptoms of frequency, urgency, haematuria, and painful micturition.^{6,7} Symptoms of bowel involvement include dyschezia, constipation, diarrhoea, or rectal bleeding, and can be wrongly diagnosed as irritable bowel syndrome (IBS).^{2,8} Catamenial complaints of chest pain, pneumothorax, haemothorax, and haemoptysis, can take place in women with thoracic endometriosis.^{9,10} Endometriosis can also occur in the abdominal wall and in scar tissue after a caesarean section or laparoscopy.¹¹ The clinical presentation is diverse, and symptoms that are exacerbated during the menstrual cycle could be a manifestation of endometriosis. The importance of the GP in the diagnosis

of endometriosis is emphasised by the fact that women with endometriosis consult primary care more frequently, offering a window of opportunity.¹²

Physical examination might reveal abnormalities (for example, pain, adnexal masses, or vaginal nodules), but normal findings do not rule out endometriosis.¹² Patients with suspected endometriosis should be referred to a gynaecologist for examination with ultrasound, MRI, or laparoscopy, because histological examination is required for the definitive diagnosis.^{3,13} In cases where symptoms are difficult to treat, a combination of hormonal and surgical resection seems to give the best outcome.¹⁴ In case of doubt, empirical management by hormonal suppression therapy with the combined oral contraceptive pill or progestins only can be started by the GP.¹ Reduced symptoms after treatment support the presumptive diagnosis of endometriosis.⁵

CONCLUSION

In conclusion, extrapelvic endometriosis is a rare presentation of endometriosis that does not have a standardised treatment protocol. In women who develop recurrent complaints with a catamenial character in the fertile age, endometriosis should always be considered. Better recognition will ultimately lead to more insight into the underlying pathology and treatment options. The importance of timely recognition and treatment is illustrated by the case presented here, with a prolonged and burdensome process that is unfortunately not uncommon for these patients.

Provenance

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