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Case report - Thoracic non-oncologic Endometriosis-related spontaneous diaphragmatic rupture

Frédéric Triponez^{a,b,*}, Marco Alifano^a, Antonio Bobbio^a, Jean-François Regnard^a

^aDepartment of Thoracic Surgery, Hôtel-Dieu Hospital and University Paris V, 1, place du Parvis de Notre-Dame, 75005 Paris, France

^bDepartment of Thoracic Surgery, University Hospital of Geneva, Geneva, Switzerland

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Abstract

Non-traumatic, spontaneous diaphragmatic rupture is a rare event whose pathophysiology is not known. We report the case of endometriosis-related spontaneous rupture of the right diaphragm with intrathoracic herniation of the liver, gallbladder and colon. We hypothesize that the invasiveness of endometriotic tissue caused diaphragm fragility, which finally lead to its complete rupture without traumatic event. The treatment consisted of a classical management of diaphragmatic rupture, with excision of the endometriotic nodule followed by medical ovarian suppression for six months.

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Keywords: Catamenial pneumothorax; Diaphragmatic rupture; Diaphragmatic hernia; Diaphragmatic repair

1. Introduction

Catamenial pneumothorax is a rare entity although recent studies suggest that it could account for up to 25% of the pneumothorax occurring in women of reproductive age [1, 2]. The classical findings at operation include mainly small diaphragmatic holes and/or endometriotic grayish nodules found preferentially in the tendinous part of the diaphragm. To our knowledge there are only two cases published in the English literature of limited diaphragmatic rupture with partial intrathoracic liver herniation probably caused by endometriosis [3, 4]. We report the case of a complete right diaphragmatic rupture associated with endometriosis with large herniation of right upper quadrant abdominal organs into the pleural cavity.

2. Case report

A 43-year-old woman presented with right chest pain in an emergency department in another hospital. She was a non-smoker and had no medical history. Physical examination showed diminished breath sounds in the upper left chest. Chest X-ray showed a right pneumothorax with elevation of the right diaphragm and pneumoperitoneum. Chest computed tomography (CT) revealed a complete liver and partial colon herniation into the chest (Fig. 1). She was in no immediate danger, was discharged home and encouraged to seek a specialized consultation.

She came to our department two weeks later. Detailed medical history revealed numerous episodes of right periscapular pain beginning the day before menstruation. She

had two children but had taken two years to get pregnant after the finishing oral contraception. She had no known history of endometriosis. At the time of admission, she complained of right chest discomfort, some dyspnea and constipation.

She was positioned in a left lateral decubitus with a double lumen endotracheal tube. The operation began with a thoracoscopic exploration showing the liver, gallbladder and colon herniation into the chest (Video 1) with a grayish nodule on a free edge of the diaphragm (Fig. 2a). A utility thoracotomy in the eighth intercostal space was performed. The herniated abdominal organs were put back in the abdomen, the endometriotic nodule was resected and the diaphragmatic rupture was repaired in a standard fashion using interrupted 1.0 absorbable suture (Fig. 2b). The suture was reinforced with a Vicryl mesh, to promote adhesions and talc poudrage was performed. Pathological examination of the resected nodule showed hemosiderin-loaded macrophages compatible with endometriosis. The postoperative course was uneventful and the patient was given ovarian suppression treatment for six months as is currently recommended in our group for catamenial pneumothorax [5].

3. Discussion

Catamenial pneumothorax is the most frequent presentation of thoracic endometriosis, which can also lead to catamenial hemothorax, catamenial hemoptysis and endometriotic lung nodule [6]. It is usually defined as the occurrence of a pneumothorax occurring in relation with the menses, 24 h before to 72 h after the onset of menses [5]. The pathophysiology of catamenial pneumothorax is not well understood but is most probably due to the passage

*Corresponding author. Tel.: +41 22 37 27 862; fax: +41 22 37 27 615.

E-mail address: frederic.triponez@hcuge.ch (F. Triponez).

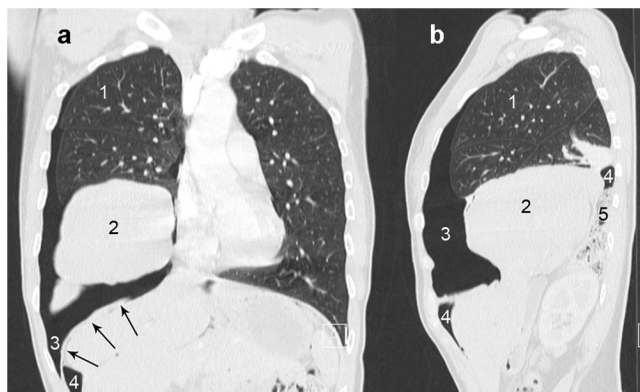
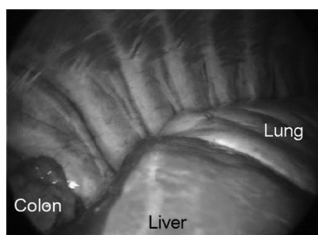


Fig. 1. Coronal (a) and sagittal (b) view of the CT. Arrows indicate the diaphragm. 1, lung; 2, liver; 3, pneumothorax; 4, pneumoperitoneum; 5, colon.



Video 1. Intraoperative view showing the intrathoracic herniation of the liver, gallbladder and right colon, with the complete diaphragmatic rupture.

of air from the abdomen through the diaphragm when there is no mucous plug in the uterine cervix. The holes in the diaphragm permitting the passage of air are explained either by congenital defects (porous diaphragm as described by Kirschner [7]) or by the invasiveness of endometriotic tissue that entered the diaphragm due to retrograde menstruations. The clockwise current of peritoneal fluid in the abdomen (down the left peritoneal gutter over the pelvic floor and up the right peritoneal gutter to the subphrenic space) together with the piston-like effect of the liver probably explains the quasi exclusively right-sided location of a catamenial pneumothorax [8]. Pathological analysis of endometriosis tissue in the pleural cavity shows stroma and glands in different proportions, almost always associated with hemorrhage, fibrosis and inflammatory infiltrates. Endometrial cells have been shown to have aggressive potential which can finally lead to involution of striated muscle fibers [9]. The periscapular pain regularly presented by our patient the day before the onset of menses could be a ‘diaphragmatic pain’, as has already described by two gynecological series on endometriosis [5].

The classical findings at operation are small holes and/or grayish-purple nodules preferentially found on the tendinous part of the diaphragm. These findings and the difficulty in visualizing them through a small axillary thoracotomy or through a thoracoscopy in the lateral position (with the arm elevated, allowing only a partial view of the diaphragm) probably explain the lack of recognition of the causative endometriosis in some of the pneumothorax, particularly in previous series.

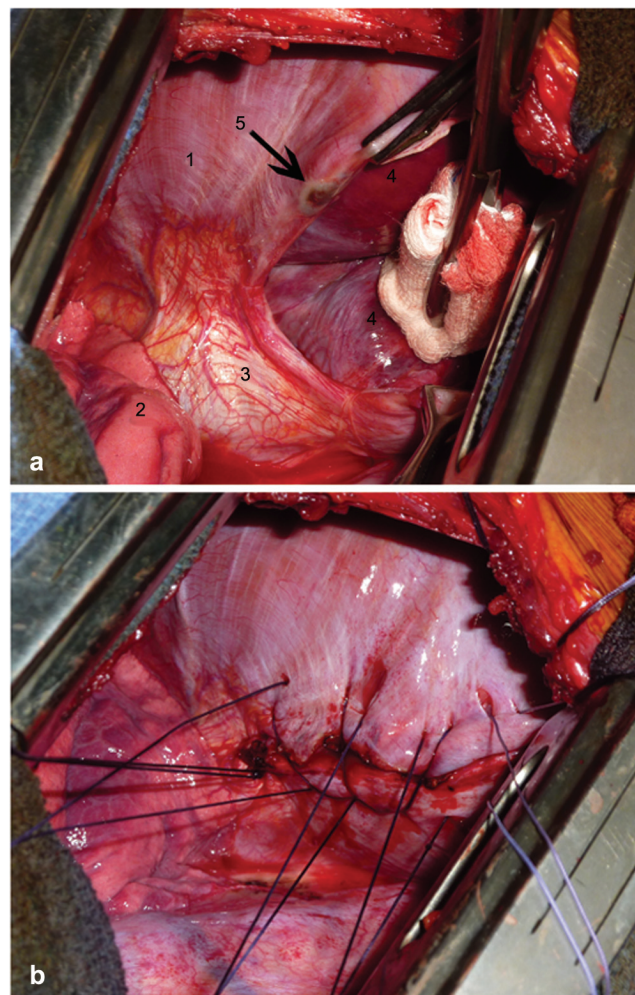


Fig. 2. Intraoperative view (a) after reduction of the liver and colon in the abdominal cavity. 1, diaphragm; 2, collapsed lung; 3, tendinous part of the diaphragm; 4, liver; 5 with arrow, endometriotic nodule and (b) after the diaphragm has been sutured.

Spontaneous diaphragmatic rupture is a rare event with only 28 reports in the English literature [10]. Most of them occurred in the left side (68% in this review [10]) which makes it unlikely that endometriosis could be the causative event in many of them. However, our report suggests that endometriosis should be searched for in patients with spontaneous diaphragmatic rupture.

In the present case, we hypothesize that the invasiveness of endometriotic tissue caused diaphragm fragility, which finally led to its complete rupture without traumatic event. The treatment consisted of a classical management of diaphragmatic rupture, with excision of the endometriotic nodule followed by medical ovarian suppression for six months. In our view, the combination of surgical and medical treatment should lead to the lower risk of recurrence of catamenial pneumothorax.

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