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Non-traumatic diaphragmatic rupture with liver herniation due to endometriosis: A rare evolution of the disease requiring multidisciplinary management

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ABSTRACT

A 35 year old woman with chronic pelvic endometriosis suffered from right scapular pain. MRI imaging showed a right diaphragmatic rupture with liver herniation. Surgical procedure was performed by thoracotomy. The liver was put back into the abdomen, endometriosis was resected from the diaphragm, interrupted non absorbable suture of the diaphragm was performed and an absorbable mesh was placed. Endometriosis was confirmed on histological analysis of the resected diaphragm. To study this pathology, we performed a systematic review of the literature and found 12 similar cases of diaphragmatic rupture due to endometriosis. Right diaphragm is often involved and rupture is always located on the tendinous portion. Symptoms are mainly cyclic right scapular pain and cathamerial pneumothorax. MRI should be performed in case of suggestive symptoms and a systematic exploration of the diaphragm should be performed at laparoscopy for an early treatment of the lesions to prevent progression to rupture.

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Introduction

Endometriosis is defined histologically by the presence of endometrial tissues (glands and stroma) outside the uterine cavity. The prevalence of endometriosis is estimated at 33% in patients with chronic pelvic pain and up to 50% in case of female infertility [1,2]. Extra pelvic involvement is rare (less than 10%) mainly affecting the gastrointestinal and urinary tract. Diaphragmatic involvement is mostly described jointly with the various thoracic disorders due to endometriosis that are grouped under the acronym TES (Thoracic Endometriosis Syndrome) associated with pelvic lesions in 84% of cases [3]. Chronic endometriosis lesions of the diaphragm can lead to diaphragmatic rupture.

We report a case of diaphragmatic rupture with liver herniation due to endometriosis in a patient with deep infiltrative pelvic endometriosis.

The patient gave her consent to participate in this case report and this study was approved by the Ethics Committee of the National College of the French Gynecologists and Obstetricians (CEROG 2018-GYN-0401).

Case report

A 35-year-old woman was referred to the department of Gynecology of the CHU of Caen (Caen, France) in March 2017 for endometriosis follow-up. She was gravida 1 para 1, and had medical history of pelvic endometriosis (stage 4) diagnosed in December 2011. She took continuous contraceptive pill during three years until right shoulder pain appeared. This shoulder pain worsened during menses and was associated with chronic pelvic pain. The patient did not present cough or cathamerial pneumothorax.

A chest X-ray showed an oval opacity of the right diaphragmatic portion without pneumothorax (Fig. 1A). A computed tomography scan without contrast injection confirmed the right diaphragmatic rupture (4.5 cm defect) with liver herniation in the pleural cavity.

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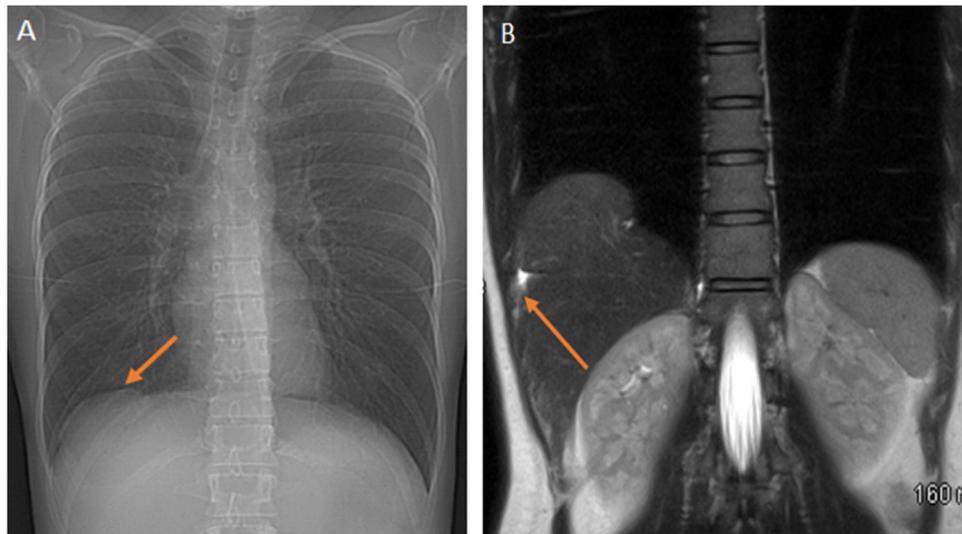


Fig. 1. A Chest X-ray: face, showing diaphragmatic opacity (yellow arrow). B MRI T2 weighted sequence showing hyper intense spots on the edge of the rupture suggesting endometriosis (yellow arrow).

Furthermore, a thickening zone was observed on the edge of the hernia, suggesting nodules of endometriosis. No other thoracic anomaly were noted on the CT-scan. MRI showed small hyperintense diaphragmatic lesions on T1 weighted images which was consistent with endometriosis. Pelvic MRI showed a 13 mm endometriotic cyst of the left ovary, a right hematosalpinx and bilateral uterosacral nodules of endometriosis (Fig. 1B).

An explorative thoracoscopy showed a 4 cm defect on the tendinous part of the right portion of the diaphragm. Typical blue-brown endometriosis lesions were observed on the edge of the rupture. A muscle-sparing mini-thoracotomy was performed on the eighth intercostal space to put back the herniated liver on abdominal cavity and to resect the endometriotic lesions. The thoracotomy was decided because of the important liver herniation with strong adherences between the diaphragm and the liver. Inspection of the abdominal side of the diaphragm (with a 30° optic) did not show other endometriosis lesions. Defect was closed with non-absorbable interrupted sutures and an absorbable mesh was placed to strengthen the diaphragm because of the large diaphragmatic defect (Fig. 2A–D). A pleural drain was placed. The patient had an uneventful recovery and pleural drain was removed at day 3 post-operative. Pathological examination was in favor of endometriosis showing glandular structures and chorion within a fibro-muscular tissue. Hormonal therapy with continuous contraceptive pill was given as patient refused GnRh analogs because of potential side effects. Six-month follow-up showed no anomaly on the chest radiography and no recurrence of shoulder pain either.

Discussion

Diaphragmatic rupture is an uncommon complication of diaphragmatic endometriosis. To specify the characteristics of this pathology we undertook a systematic literature review. The following descriptors were used on PubMed/MEDLINE database: “diaphragmatic endometriosis”, “diaphragmatic rupture”, “thoracic endometriosis”. We identified 12 cases reported, considering only the articles in English [4–10]. Characteristics of the 12 cases are reported in the Table 1.

The case we report has many similarities with the 12 cases of the literature. The patient had a chronic pelvic endometriosis and she was suffering from right shoulder pain. Her symptomatology was poor considering the important diaphragmatic defect she presented. However, the surgery was indicated because the

shoulder pain worsened and to avoid widening of the rupture and liver herniation with potential impact on respiratory functions. In case of diaphragmatic endometriosis the symptomatology is poor with symptoms in only 30% of the cases, nevertheless, right scapular pain and recurrent cathamerial pneumothorax are common signs of diaphragmatic rupture requiring thoracic imaging [3,11].

In our case, chest radiography using two incidences showed the diaphragmatic anomaly. A CT-scan is often performed as a first-line examination for thoracic screening. It allows the assessment of the rupture, its dimensions and if there are any herniated organs. Diaphragmatic MRI brings more specific signs for the etiology of rupture. Indeed, the presence of pleural thickenings, hyper intense in T1 and T2 weighted sequences is suggestive of nodules of endometriosis (and even more when pelvic endometriosis is associated). Rousset et al, showed that MRI had a 83% sensitivity to diagnose diaphragmatic endometriosis [12]. Diaphragmatic MRI should be performed when diaphragmatic endometriosis is suspected.

The physiopathology of diaphragmatic endometriosis remains unclear. Numerous series of thoracic endometriosis highlight that pelvic endometriosis seems to be strongly associated [1]. Moreover, there might be a chronological evolution from pelvic involvement to the occurrence of diaphragmatic endometriosis, with an average delay of 10 years [13]. Diaphragmatic endometriosis is therefore probably due to peritoneal dissemination of endometrial tissues from menstrual reflux or pelvic endometriotic tissue would migrate through the right paracolic gutter to the right hypochondrium and would be trapped by the falciform ligament [14]. Thus, chronic reflux of endometrial tissue trapped in right hypochondrium would result in numerous diaphragmatic involvement.

Diaphragmatic ruptures due to endometriosis are more often localized in the tendinous portion of the diaphragm. This thick portion might be easily involved and became porous because of deep infiltrative endometriosis. Inflammatory lesions linked to endometriosis can lead to confluent defects and rupture with organ herniation in the pleural cavity due to abdominal pressure. Furthermore, these defects could create diaphragmatic pathways, ectopic implants could go through to involve the pleural cavity.

It should be noted that spontaneous rupture of the diaphragm has been reported without history of endometriosis, it could be due to preexisting diaphragmatic fragility or malformation. These

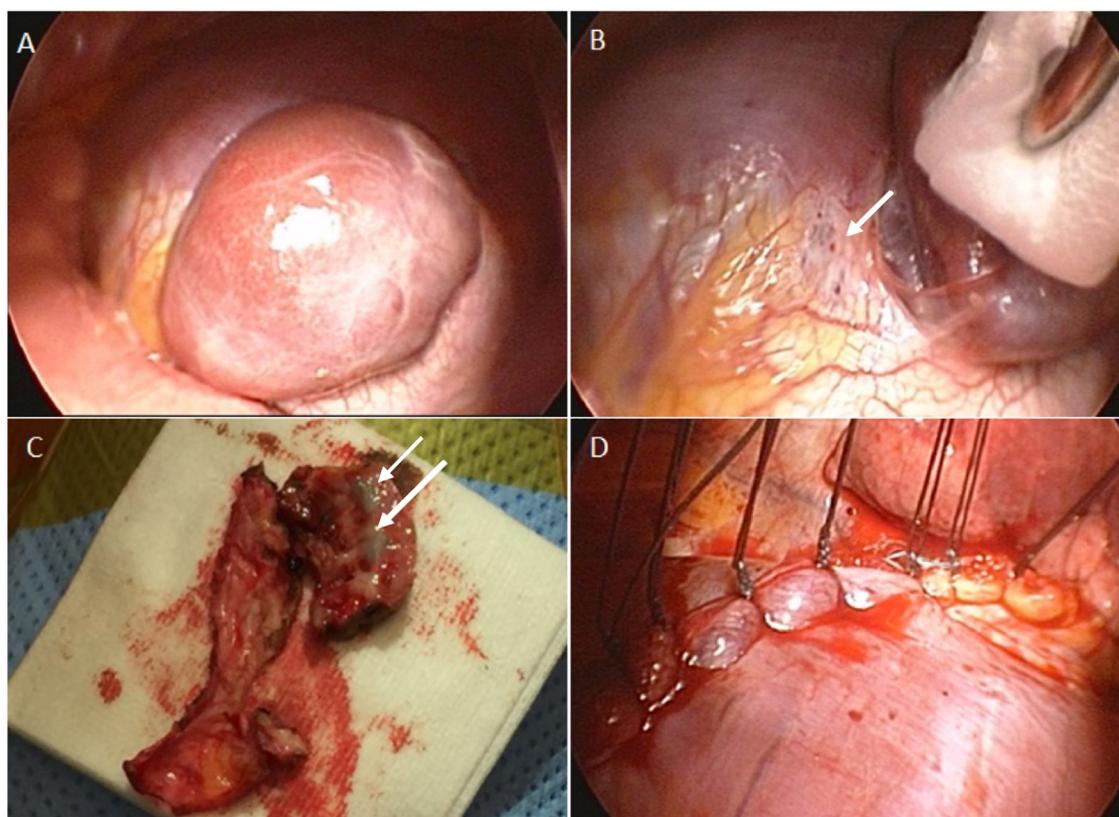


Fig. 2. A Thoracotomy view, diaphragmatic defect with liver herniation. B Endometriosis spots on the tendinous portion of the diaphragm (white arrow). C Resected diaphragm, endometriosis with typical blue-brown color (white arrow). D Defect closed with non-absorbable interrupted sutures.

spontaneous ruptures are left-sided in 68% of the cases and localized in the peripheral diaphragmatic portion in 79% of the cases ; instead in case of endometriosis, lesions are typically right sided, in central position [15].

Surgical management is always necessary in case of diaphragmatic rupture. We found that eleven reported cases had a video assisted thoracoscopy and one had a thoracotomy. In our case, the surgical management of the rupture can not be led by a total laparoscopic approach due to difficulty of exposition on the posterior

portion of the diaphragmatic rupture, furthermore, the thoracoscopy allowed the inspection of the pleural cavity and the dissection of pleural adhesences which were to strong and lead to conduce a thoracotomy for more security. The excision of endometriotic lesions followed by non-absorbable interrupted suture is often performed. An absorbable mesh can be used in case of important defect and talc pleurodesis could be benefic in case of cathamential pneumothorax history. Hormonal therapy for ovarian suppression might be helpful to slow down endometriosis recurrence.

Table 1
Characteristics of the 12 cases of diaphragmatic rupture due to endometriosis.

First author	Year	Case N°	Age	Medical background	Clinical features	Lesions	Side	Associated herniation	Management	Recurrence
Bobbio [2]	2007	1	35	3 CPN	Recurrent CPN	Tendinous rupture	Right	Liver	Thoracoscopy Interrupted suture	NA
Triponez [3]	2010	2	43	No	Right chest pain	Tendinous rupture	Right	Liver Colon	Thoracotomy Interrupted suture + absorbable mesh	NA
Haratake [4]	2015	3	50	Chilaiditi syndrome	Small bowel obstruction	Diaphragmatic rupture	Right	Small bowel	Thoracoscopy and laparotomy	NA
Pryshchepau [5]	2010	4	42	NA	Right PN	Tendinous rupture	Right	Liver	Thoracoscopy	NA
Makhija [6]	2012	5	38	Pelvic endometriosis	Right CPN	Tendinous rupture	Right	No	Thoracoscopy Diaphragm plicature + talc pleurodesis	No
Bobbio [7]	2017	6-11	29-50	Diaphragmatic endometriosis (4) No (2)	Scapular pain (6)	Diaphragmatic rupture	Right (5) Left (1)	Liver (5) Colon (1)	Thoracoscopy (6) Interrupted suture (6), mesh (2)	NA
Afzal [8]	2016	12	39	CPN	Right chest pain	Diaphragmatic rupture	Right	Liver	Thoracoscopy pleurectomy and pleurodesis	NA

CPN: cathamential pneumothorax, PN: pneumothorax, NA: not available data.

To conclude, diaphragmatic endometriosis is rare but can progress to diaphragmatic rupture. It is mostly right sided and often involves the tendinous portion of the diaphragm. The symptoms are rather subtle and do not foreshadow the importance of diaphragmatic defect. Deep pelvic endometriosis is usually associated and precedes diaphragmatic rupture by several years. Diaphragmatic MRI is the key examination to diagnose rupture and its origin with fat-suppressed T1-weighted sequences. A multidisciplinary approach is necessary to choose the best therapeutic option. Surgical management of the diaphragmatic rupture can be led by thoracoscopy. Thoracotomy might be useful in cases of complex adhesions between the diaphragm or lung and herniated organs. Excision of endometriosis on the edge of the hernia can make the defect wider and requires the placement of a mesh.

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