

## Laparoscopic management of diaphragmatic endometriosis: what can we learn

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**SUMMARY:** Laparoscopic management of diaphragmatic endometriosis: what can we learn.

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**Study Objective.** To describe the diagnosis, surgical treatment and long term symptomatic outcome of diaphragmatic endometriosis.

**Patients and methods.** Nine consecutive women with diaphragmatic endometriosis were identified in a retrospective review of women undergoing laparoscopic surgery for pelvic endometriosis, between September 2005 and September 2012. Symptoms suggestive of diaphragmatic endometriosis in a patient with pelvic disease were considered. The diagnosis was based on the characteristics of endometriotic lesions observed during laparoscopic session.

**Results.** Operative findings in 9 patients, included 2 superficial nodules of endometriosis on the diaphragm measuring about 1 cm. Seven women had numerous lesions scattered across the diaphragm. Lesions were bilateral in 2 patients, limited to the right hemidiaphragm in 6 patients, and limited to the left hemidiaphragm in 1 patient. The atypical symptoms in 2 symptomatic patients decreased significantly after treatment, with a minimum follow-up period of 12 months. No postoperative complications occurred.

**Conclusions.** The diaphragmatic endometriosis can be diagnosed and treated effectively with the use of laparoscopy, when the lesions are superficial and in the safe area.

**RIASSUNTO:** Il management laparoscopico dell'endometriosi diaframmatica: cosa possiamo imparare.

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**Obiettivo.** Descrivere diagnosi, trattamento e outcome clinico del trattamento laparoscopico dell'endometriosi diaframmatica.

**Materiali e metodi.** Attraverso un'analisi retrospettiva dei casi di endometriosi pelvica trattati nel periodo compreso tra settembre 2005 e settembre 2012, sono stati identificati nove casi consecutivi di endometriosi diaframmatica trattata per via laparoscopica.

Abbiamo registrato tutti i sintomi atipici riferiti, suggestivi di localizzazione diaframmatica dell'endometriosi. La diagnosi è stata effettuata sulla base delle caratteristiche delle lesioni osservate durante il tempo operatorio.

**Risultati.** Delle 9 pazienti con lesioni endometriose diaframmatiche, in due casi abbiamo identificato la presenza di noduli diaframmatici superficiali, di circa 1 cm di diametro. Sette donne presentavano numerose lesioni di piccolo diametro sparse sulla superficie diaframmatica. Le lesioni erano bilaterali in 2 casi, limitate all'emidiaframma destro in 6 casi, e limitate all'emidiaframma sinistro in 1 solo caso. I sintomi atipici riscontrati in 2 pazienti in fase pre-operatoria si sono ridotti in modo significativo dopo il trattamento, con un follow-up minimo di 12 mesi. Non si sono verificate complicanze post-operatorie.

**Conclusioni.** Le lesioni endometriose del diaframma possono essere diagnosticate e trattate per via laparoscopica in modo efficace, se superficiali e in un'area sicura.

**KEY WORDS:** Diaphragmatic endometriosis - Laparoscopy - Extragenital endometriosis. Endometriosi diaframmatica - Laparoscopia - Endometriosi extragenitale.

## Introduction

Approximately 8 to 15% of the female population of reproductive age is affected by endometriosis, that is usually confined to the pelvis. Sometimes endometriotic lesions are localized out of the pelvis and in the diaphragm. Diaphragmatic involvement is a rare condition, affecting from 0,6 to 1,5% of patients undergoing surgery for endometriosis (1, 2). Clinical findings of diaphragmatic endometriosis are often non-specific or absent mainly because lesions are often superficial. However, in a patient affected by pelvic endometriosis, chest, hypochondrium or shoulder catamenial pain (with arm or cervical irradiation) strongly suggests a diaphragmatic involvement (1, 3).

Despite, the etiology of endometriosis is still controversial, the diaphragmatic endometriosis may shed light on its pathogenesis, since the asymmetric distribution of diaphragmatic endometriotic lesions seems supporting the menstrual reflux theory (4).

This article describes the author's experience on diagnosis, treatment, and symptoms response in nine women with diaphragmatic endometriosis and focuses on incidence of diaphragmatic endometriosis and pathogenesis.

## Method and materials

Nine consecutive women with diaphragmatic endometriosis were identified in a retrospective review of women undergoing to laparoscopic surgery for pelvic endometriosis at the University Hospital "P. Giaccone" in Palermo, between September 2005 and September 2012.

Pre-operative evaluation included patient's history and clinical and instrumental assessment (5). Symptoms hinting diaphragmatic endometriosis, in a patient with pelvic disease, were considered dull shoulder tip pain, lateral and anterior thoracic pain or hypochondrium pain irradiating to the arm or cervical region and worsening during menses. All patients underwent an ultrasound assessment by transvaginal and transabdominal approach. Other imaging evaluations (magnetic resonance, double-contrast barium enema, computerized tomography) were performed only when deep infiltrating endometriosis, intestinal or extra-peritoneal diseases were suspected.

Diagnosis was based on specific characteristics of endometriotic lesions observed during laparoscopic session. The staging of the endometriosis was carried out using the procedure specified by the Revised American Fertility Society (r-AFS) (6).

The median age of the patients was 32 years (range, 22-41 years), out of which 2 had been pregnant previously. The main indication for laparoscopy in all the women was pelvic pain (dysmenorrhea, dyspareunia). A total of 3 patients presented with symptoms highly suggestive of diaphragmatic endometriosis, including shoulder pain, chest pain, and right upper quadrant pain. Two patients underwent previous surgery for pelvic endometriosis (ovarian endometriotic cyst, in both cases), by laparoscopy and by laparotomy respectively. One patient received 2 previous laparoscopic surgery.

All patients were instructed routinely on: the possibility that endometriosis involving non-pelvic organs, such as the urinary tract, intestine, or diaphragm, might be discovered; the intent of treating such endometriosis; the possible complications of this type of surgery. A formal consent form was obtained from all patients.

All procedures were performed in our referral center by expert surgeons, under general endotracheal anesthesia. The laparoscope was inserted from a 10 mm umbilical port and three 5 mm port were then placed on each side and centrally, under direct vision (7).

The diaphragm dome was carefully examined as part of the routine abdominal exploration, at the beginning of each laparoscopic procedure. When needed, a more accurate exploration of the diaphragm was obtained by placing the patients in a steep reverse Trendelenburg position or using a 30° optics. Similarly to the arrangement for laparoscopic cholecystectomy or splenectomy (8-10), when diaphragmatic endometriosis was noted and surgical treatment was necessary, additional trocars were placed in the upper right or left quadrant according to the implant location. A blunt probe was used to retract the liver inferiorly and better expose the posterior area of the diaphragmatic dome.

Lesions were classified in peritoneal foci, usually less than 0.5 cm in diameter, superficial nodules, superficial lesions of 0.5-1 cm of diameter, and infiltrating nodules, usually greater than 1 cm and penetrating the diaphragmatic muscularis.

Peritoneal foci has been treated only in symptomatic patients, in which lesions were easy to access and far from cardiac area or phrenic nerve. Superficial nodules has been treated if located at a safe distance from left ventricle and/ or phrenic nerve.

Technical difficulty and risk of perforating diaphragm discouraged doing routinely biopsies. However, in all cases, histological examination of specimens from other pelvic or abdominal lesions confirmed endometriosis. All patients presented en-

dometriosis disease at other sites, which was handled in the same session. All operations were always documented by video.

Patients diagnosed with a III and IV AFS-r stage underwent a GnRH-analogous therapy for 6 months after surgery and then continued with an estro-progestinic regimen. Patients were followed-up by interview, clinical evaluation and ultrasound control at 1 month, 6 months and 12 months after surgery.

## Results

On 986 patients treated by laparoscopy for pelvic endometriosis, in 9 cases (0,9%) a diaphragmatic involvement were identified, during the study period (Table 1).

Five patients (55,5%) displayed small peritoneal foci (from 1 to 6) of diaphragmatic endometriosis measuring < 0,5 cm in diameter; 2 cases showed numerous peritoneal foci (> 6), scatteredly distributed on the diaphragm surface; 2 women presented one superficial nodule (Figure 1), in one case associated to scattered superficial foci. The lesions were bilateral in 2 patients, limited to the right hemidiaphragm in 6 patients and localized on the left hemidiaphragm just in one patient (Table 1).

The endoscopic appearance of the lesions was variable: smooth red blebs with surrounding angiogenesis, flat whitish stellate lesions and hemosiderin pigmented dark lesion of variable diameter (Figure 1). In no case, endometriosis was observed to deeply infiltrate the muscular layer of the diaphragm.

Superficial foci were treated by bipolar electrocoagulation in symptomatic patients. The only case of left peritoneal lesion was not cured, since it was very close to the left ventricle and it was asymptomatic. In 2 cases of bilateral lesions, only the right hemidiaphragm ones in the symptomatic patients were handled (case n.3). One superficial asymptomatic nodule was fulgurated, since it was an isolated lesion, it was located on right hemidiaphragm and a minimal lesion was found over hepatic glissonian (case n.6).

The total of 9 patients had pelvic endometriosis and endometrial implants also were found on the bladder (case n.7) and on the bowel (cases n.3 and n.7). In 2 cases we performed endometrial polypectomy by hysteroscopy in the same surgical session (11). Two patients have been described as stage IV, 6 as stage III and just one case as stage II of AFS-r. Operating time was 90 minutes (range 40-140 minutes), while the treatment of diaphragmatic endometriosis required 10 minutes (range 5-20 minutes).

Neither intra-operative complications nor any post-operative complications related to the treatment of the diaphragmatic lesions occurred. Patients were discharged after 3 days of hospitalization. Three symptomatic patients experienced significant reductions in diaphragmatic endometriosis pain, within a 12 months follow-up. One symptomatic patient in which lesions were treated partially on the left side of the diaphragm (case n.3) started complaining about a chest pain on the left side, 10 months after surgery.

TABLE 1 - CLINICAL AND SURGICAL CHARACTERISTICS OF PATIENTS WITH DIAPHRAGMATIC ENDOMETRIOSIS.

Pt	Age	Symptoms	AFS-r Stage	Type	n. lesions	Pelvic endometriosis	Side	Treatment
1	29	no	III	peritoneal foci	5	yes	right	no
2	35	no	II	peritoneal foci	4	yes	right	no
3	41	yes	IV	superficial nodule	>6	yes + bowel	bilateral	electrocoagulation
4	23	yes	III	diffuse peritoneal foci	>6	yes	right	electrocoagulation
5	40	no	III	peritoneal focus	1	yes	left	no
6	32	no	III	superficial nodule	1	yes + glissonian	right	electrocoagulation
7	33	yes	IV	diffuse peritoneal foci	>6	yes + bladder + bowel	right	electrocoagulation
8	27	no	III	peritoneal foci	4	yes	bilateral	no
9	22	no	III	peritoneal foci	2	yes	right	no

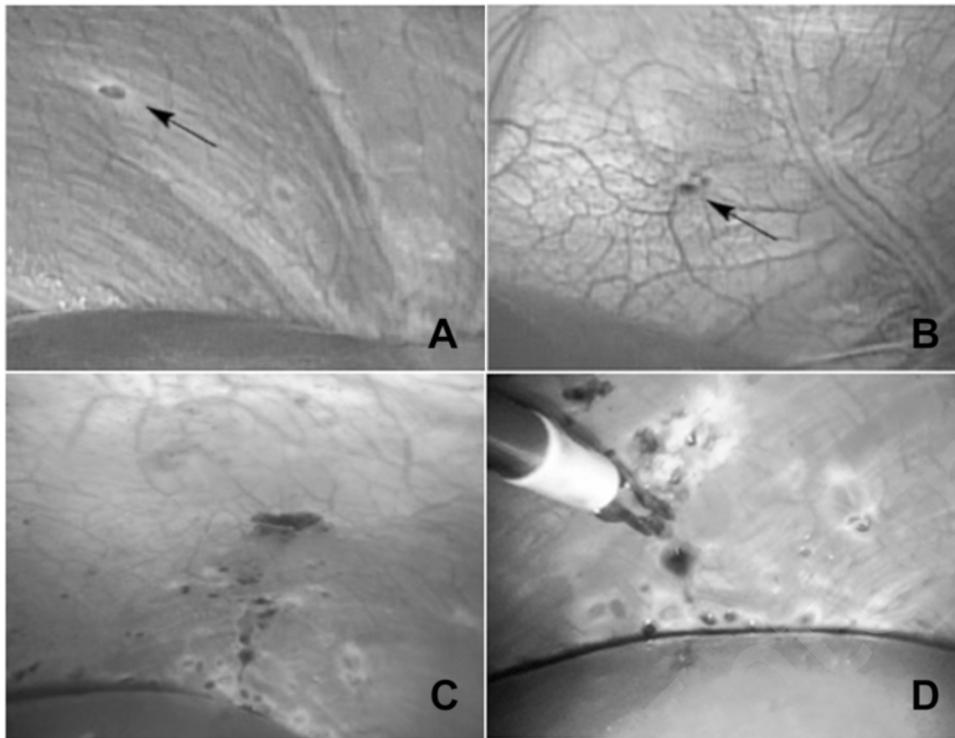


Figure 1 - Peritoneal foci of endometriosis in the left (A) and right (B) hemidiaphragm. Diaphragmatic superficial nodules and scattered peritoneal foci of endometriosis (C-D).

## Discussion

Endometriosis is an interesting chronic disease defined as the presence of endometrial tissue outside of the uterus in lesions of variable sizes and appearances (12). Endometriosis may be both asymptomatic or associated with symptoms of pain and/or infertility (13).

Endometriosis usually affects pelvic organs, however it may spread to the upper abdomen, from 0,6% (14, 15) to 12% of cases (16). Diaphragmatic endometriosis may constitute a serious issue, as it may cause severe pain syndromes and it may extend to contiguous organs, such as liver, pleura and lung.

Among patients undergoing surgery for endometriosis, an incidence ranging of 0,19% has been formerly reported (17). In our series, we had an incidence of 0,9%, which is midway between previous data and more recent findings of Ceccaroni et al. (2). We suppose that the higher incidence values documented may be explained by the greater accuracy in the inspection of the diaphragm, both in its posterior leaf and right lateral wall and in the falciform ligament (4).

Several considerations arise from the analysis of the data reported in this series. First, the majority of

patients with diaphragmatic endometriosis do not complain about it. In our series, only 3 patients out of 9 referred pain to their chest, upper abdominal quadrants, shoulder or arm, likely because of phrenic nerve irritation. As well as diaphragmatic endometriosis, any superficial lesion occurring in any area of the pelvis may be asymptomatic. For example, superficial endometriosis can occur both on the bowel and the bladder, without producing intestinal or urinary symptoms (18).

In our series, diaphragmatic endometriosis was found mostly in women with severe pelvic endometriosis (III-IV stage AFS-r). This may be related to the etiologic mechanism of this disease. Unequivocal pathogenesis of endometriosis is still unknown, but the most widely accepted theory relates it to retrograde menstruation through the fallopian tubes with subsequent dissemination of endometrial cells within the peritoneal cavity (19). Such etiology seems to be corroborated both by the constant association between endometriotic pelvic disease and diaphragmatic involvement and by the asymmetric distribution pattern of endometrial diaphragmatic implants. A clockwise intraperitoneal flow would transport endometrial refluxed cells to the upper right abdominal quadrant. Here, the falciform liga-

ment of the liver would prevent their passage to left quadrant, thus making right diaphragmatic implants significantly more frequent than left ones (4).

Taking into account the limited role of diagnostic imaging, diagnosis of asymptomatic diaphragmatic endometriosis before explorative surgery is difficult. Since an high degree of suspicion is needed, patients should be asked for symptoms suggestive of diaphragmatic involvement (18). Catamenial pneumothorax is a rare and severe complication of infiltrating diaphragmatic endometriosis, which, although highly indicative, did not occur in any of our patients.

Even though diaphragmatic lesions may spread to adjacent organs with increased morbidity, the natural history of diaphragmatic endometriosis is unknown and its surgical treatment may expose patients to various complications. For these reasons, proposing surgical recommendations is difficult in such situations, but we suggest treatment of symptomatic patients. Deeply infiltrating nodules should be resected, however the support of a thoracic sur-

geon is needed, as the approach is difficult and a complete removal is necessary.

Moreover, patients should be preoperatively, adequately advised about the risks related to the extensive treatment of endometriosis affecting extrapelvic organs and should receive one or more cycle of GnRH-analogous therapy after surgery. The treatment of asymptomatic lesions, in order to prevent progression and spread to other organs, relies both on the intraoperative evaluation of diaphragmatic lesions and on the features of the pelvic involvement. Since diaphragmatic endometriosis is an operative diagnosis, surgical strategy is decided during surgery. We approached superficial endometriotic lesions involving the diaphragm when measuring less than 0,5 cm in diameter and if are located in a safe area, as electro-surgery should not be used close to the left ventricle, due to undesired heat transmission. Finally, since laparoscopy allows an easy and thorough exam of the entire abdominal cavity, including the diaphragm, visual inspection should be performed routinely when dealing with endometriosis.

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