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# Laparoscopic surgical management of diaphragmatic endometriosis :

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Objective: To review the clinical presentations of and management options for diaphragmatic endometriosis.

Design: Retrospective review.

Setting: Referral center.

Patient(s): Twenty-four women with endometriosis of the diaphragm.

Intervention(s): Surgical management.

Main Outcome Measure(s): Diagnostic accuracy and therapeutic feasibility of operative laparoscopy.

Result(s): Operative findings in 17 patients included 2-5 spots of endometriosis on the diaphragm measuring <1 cm. Seven women had numerous lesions scattered across the diaphragm. Lesions were bilateral in 8 patients, limited to the right hemidiaphragm in 14 patients, and limited to the left hemidiaphragm in 2 patients. In 7 patients, six endometriosis lesions were directly in the line of the left ventricle and three lesions were adjacent to the phrenic nerve. Endometriosis was infiltrating into the muscular layer of the diaphragm in 7 patients. The symptoms in all 7 symptomatic patients decreased significantly after treatment, with a minimum follow-up period of 12 months. No postoperative complications occurred.

Conclusion(s): The abdominal diaphragm can be involved with endometriosis and can be diagnosed and treated effectively with the use of videolaparoscopy. (Fertil Steril<sup>®</sup> 1998:69:1048-55. ©1998 by American Society for Reproductive

Key Words: Laparoscopy, diaphragmatic endometriosis, extragenital endometriosis

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Endometriosis has been observed in 8%-15%of women of reproductive age (1), and its involvement in nonpelvic organs is not uncommon (2). Diaphragmatic endometriosis has been the subject of incidental case reports (3-6). It may be asymptomatic, produce pain in the right or left upper quadrant of the abdomen, or refer pain to the right or left shoulder. When there has been penetration of the diaphragm with extension into the pleural space, pneumothorax or hemothorax, followed by hemoptysis, may result. This rare condition raises several diagnostic and treatment challenges. We present a series of patients whose conditions were diagnosed as diaphragmatic endometriosis and describe the clinical presentation of the disease and the surgical options for its management.

## MATERIALS AND METHODS

Twenty-four women with diaphragmatic endometriosis were identified in a retrospective chart review of women with endometriosis who were treated at the Center for Special Pelvic Surgery in Atlanta, Georgia, or at the Institute for Special Pelvic Surgery in Palo Alto, California, between April 1990 and November 1996. The diagnosis was based on characteristic endometriosis lesions observed during videolaparoscopy. Patients with suspicious lesions such as hemosiderin deposits were not included.

The median age of the patients was 31 years (range, 18-43 years). Only 7 of the women previously had been pregnant, I twice. The main indication for laparoscopy in all the women was pain. A total of 7 patients presented with symptoms highly suggestive of diaphragmatic endometriosis, including shoulder pain, chest pain, and right upper quadrant pain (Table 1). One woman had cyclic epigastric pain related to menses (case 2). During the initial patient interview, none of the remaining

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17 patients reported any symptoms that could be related directly to the diaphragmatic endometriosis.

Pelvic endometriosis previously had been diagnosed in all but 4 of the patients. Of the remaining 20 patients, endometriosis was diagnosed in 1 (case 1) after biopsy of a vaginal lesion and in the other 19 either by laparotomy or by laparoscopy. These 19 patients had undergone a median of one previous surgery (range, 1-4 surgeries) for the diagnosis or treatment of endometriosis. Eighteen of them also previously had received different types of hormone suppression therapy.

The 7 symptomatic patients who were suspected of having diaphragmatic endometriosis were prepared for treatment before surgery (cases 1–7). Fourteen patients had superficial endometriosis diagnosed during laparoscopy and were treated. Three asymptomatic patients (cases 22–24) were not treated because a thoracic surgeon was not available.

All the patients were instructed routinely regarding the possibility that endometriosis involving nonpelvic organs such as the urinary tract, intestine, or diaphragm would be discovered, as well as the intent to treat such endometriosis and the possible complications of this type of surgery. A formal consent form was obtained from all patients, without specific institutional review board approval, because the patients described were not enrolled in a research protocol and surgical management of endometriosis involving multiple organs is routine.

Laparoscopy was performed in a tertiary care center, through an umbilical site in the manner previously described (7). When endometriosis infiltrating the muscularis of the diaphragm was encountered unexpectedly, its treatment was not begun until it had been verified that a thoracic surgeon was immediately available if needed.

All procedures were performed under general endotracheal anesthesia with the patients in a modified dorsolithotomy position. The diaphragm was examined as part of the routine abdominal exploration completed during the initial phase of every laparoscopic procedure. If better visualization of the diaphragm was required, the patients were placed in a steep reverse Trendelenburg position. When endometriosis involving the diaphragm was strongly suspected or when surgical treatment was needed after the diagnosis was made, additional trocars were placed in the upper right or left quadrant according to the implant location, similar to the arrangement for laparoscopic cholecystectomy or splenectomy. An atraumatic grasping forceps or suction irrigator probe was used to push the liver down and away from the operative field and provide better exposure of the diaphragm.

When the lesions were superficial (only serosal surface involvement without penetration to the muscularis), they were vaporized using a combination of hydrodissection and a carbon dioxide (CO<sub>2</sub>) laser (15–25 W) or the Cavitational Ultrasonographic Surgical Aspirator (CUSA: Valleylab.

Boulder, CO). Hemostasis was achieved with bipolar electrocoagulation.

Deep endometriosis lesions infiltrating the diaphragm muscularis were removed with the use of hydrodissection and sharp dissection with the aid of the CO<sub>2</sub> laser set at 20–25 W (Figs. 1–3). A laparoscopic aspirating needle was inserted through one of the ancillary ports and used to inject 5–10 mL of lactated Ringer's solution under the lesions. The lesions were grasped with a grasping forceps and the CO<sub>2</sub> laser was used to make an incision around them. While the suction irrigator probe was used for traction, evacuation of smoke, and as a backstop for the CO<sub>2</sub> laser, the lesions were excised. Any remaining lesions were vaporized.

Biopsies were not performed routinely because of technical difficulty and danger of perforating the diaphragm. However, in all cases, endometriosis was confirmed histologically in other sites of the pelvis or abdomen. All the women had involvement of different pelvic organs with endometriosis, which was treated. Diaphragmatic endometriosis was documented photographically in all patients.

#### **RESULTS**

The operative findings in 17 patients included 2–5 small spots of diaphragmatic endometriosis measuring <1 × 1 cm in diameter. The remaining 7 women had numerous lesions scattered on the diaphragm (Fig. 1). The lesions were bilateral in 8 patients, limited to the right hemidiaphragm in 14 patients, and limited to the left hemidiaphragm in 2 patients. In those patients who had bilateral involvement, the lesions generally were spread more densely on the right side. In 7 patients, six endometriosis lesions were noted to be directly in the line of the left ventricle and three were adjacent to the phrenic nerve. Endometriosis was observed to be deeply infiltrating into the muscular layer of the diaphragm in 7 patients, 4 of whom were symptomatic.

All but 5 of the patients had extensive endometriosis of the pelvis. Endometrial implants also were found on the ureter (n = 9), the bladder (n = 13), the bowel (n = 16), and the appendix (n = 9). Additional findings included complete obliteration of the cul-de-sac (n = 9) and bilateral endometrioma (n = 6).

In 3 patients, the endometriosis lesions on the diaphragm were not treated. In 20 patients, the lesions were treated by CO<sub>2</sub> laser ablation, and in 1 patient, the CO<sub>2</sub> laser and CUSA were used. In the last patient (case 2), sporadic premature ventricular contractions occurred during the treatment of endometriosis in the left hemidiaphragm while the CUSA was being used. No other intraoperative complications occurred. There were no postoperative complications related to the treatment of the diaphragmatic endometriosis. All 7 symptomatic patients experienced significantly reduced diaphragmatic endome-

### Characteristics of 24 women with diaphragmatic endometriosis.

Case	Symptoms of diaphragmatic endometriosis	Stage of pelvic endometriosis	Severity and location of diaphragmatic endometriosis	Type of treatment	Duration of follow-up (mo)	- Follow-up results
1	Chest pain, RUQ pain, severe upper abdominal pain	ĮV	Superficial: bilateral, scattered	CO <sub>2</sub> laser	. 17	Pain and symptoms decreased after surgery
2	Cyclic epigastric and mid-upper	1	Deep: two spots on left side in line with left ventricle	CUSA	50	Doing well: receiving BCP
3	abdominal pain Right shoulder pain	IV	Superficial and deep: scattered, but mainly on the right side, also around	CO <sub>2</sub> laser	49	Underwent laparoscopy with LAVH/RSO and bowel resection 6 weeks later; doing well 4 y
4	Chest, RUQ shoulder	I	phrenic nerve and left ventricle Superficial and deep: five	CO <sub>2</sub>	12	later  Significant pain relief after the first
	pain		spots on the right side, including one bordering the phrenic nerve	luser		procedure for 1 y, but the pain gradually returned; after medical therapy failed, she underwent a
÷	· · · · · · · · · · · · · · · · · · · ·					second operative laparoscopy 68 mo later with significantly reduced diaphragmatic endometriosis pain
5 .	Chest, RUQ, shoulder pain	II	Deep: bilateral, scattered on the right side and one spot on the left side over the	CO <sub>2</sub> laser	29	Significantly decreased pain after surgery, but the pain returned after 2 y; she underwent another
			heart and left ventricle			laparoscopic surgery for the treatment of pelvic pain and no endometriosis of the diaphragm was seen. As of this writing, she
6	RUQ/right-sided chest	IV	Superficial: two spots on the right side	CO <sub>2</sub>	27	was trying to conceive Significant reduction in pain
7	RUQ pain	IV .	Superficial: scattered on the right side, one spot on the left side	CO <sub>2</sub> laser	16	Significant reduction in diaphragmatic symptoms; pelvic pain currently is being managed
8	, None	IV	Superficial: one spot on each side, including one in line with the left ventricle	CO <sub>2</sub> laser	29	with a GnRH analogue Pain reduced by >50%, and became pregnant with IVF after 2 y
9	None	IV	Superficial: scattered on the right side, around the phrenic nerve	CO <sub>2</sub> laser	72	Two subsequent laparoscopies for pelvic pain/endometriosis; no diaphragmatic endometriosis found
10	None	IV	Superficial: two spots on the right side	CO <sub>2</sub> laser	38	Doing very well: achieved pregnancy spontaneously and gave birth to twins by cesarean section
11	None	IV	Superficial and deep: twenty spots on the right side and one spot on the left side	CO <sub>2</sub> laser	. 3	Doing well: receiving BCP
12	None ;	IV .	Superficial: two spots on the right side	CO <sub>2</sub> laser	75	Laparoscopy performed 6 y later: two new spots found on the right side, as well as the mild adhesions from the previous
3	None	IV	Deep: one large spot on the right side	CO <sub>2</sub> laser	12	treatment  Doing well: became pregnant
4	None	IV [	Superficial and deep: ten spots on the right side	CO <sub>2</sub>	10	Doing well: receiving BCP
5	None	IV	Superficial: three spots on the right side, one spot on the	CO <sub>2</sub> laser	7	Doing well: receiving BCP
6	None	IV	left side Superficial: scattered on the right side	CO <sub>2</sub> laser	14 15	Doing well: no pain
7	None	IV	Superficial: two spots on the right side	CO <sub>2</sub> laser	2	Doing well; no pain

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eristics of 24 women with diaphragmatic endometriosis.

Symptoms of diaphragmatic endometriosis	Stage of pelvic endometriosis	Severity and location of diaphragmatic endometriosis	Type of treatment	Duration of follow-up (mo)	Follow-up Tresults
None	IV	Superficial: two spots on the	CO <sub>2</sub>	4	Doing well: no pain
None	II	right side Superficial: two spots on the right side	laser CO <sub>2</sub> laser	2	Still experiencing rectal pain that started before surgery; receiving BCP
None	1V	Superficial: two spots on each side	CO <sub>2</sub> laser	21	Doing well with no pain; trying to become pregnant
None	IV	Superficial: scattered on the right side	CO <sub>2</sub> laser	48	Doing well: underwent two subsequent laparoscopic surgeries, including one for
	•		•		pelvic pain, pelvic endometriosis, and ureteral stricture, and another for ovarian remnant syndrome and
		:			adhesions: there was no sign of diaphragmatic endometriosis at
None	IV	Superficial: one spot on the right side	None	12	either surgery Pain is manageable; trying to become pregnant
None	IV.	Appeared superficial: four spots on the right side	None	26	Doing well; became pregnant, but had a subsequent miscarriage
None		Appeared superficial: two spots on the left side over the left ventricle	None	27	Became pregnant; during her pregnancy she became symptomatic with severe left- sided chest pain; the pain
					worsened after delivery and discontinuation of breast-feeding; all other work- ups were negative

<sup>=</sup> birth control pills; CUSA = cavitational ultrasonographic surgical aspirator; LAVH = laparoscopically assisted vaginal hysterectomy; RSO = 120-00phorectomy; RUQ = 120-00pho

ymptoms, with a minimum follow-up period of 12

symptomatic patient with deep diaphragmatic endos (case 4) had significantly reduced pain in her chest, per quadrant, and shoulder for >1 year. However, gradually returned. After a trial of different types of suppression therapy, she eventually underwent anocedure almost 6 years later. At that time, the endos was resected and she, experienced reduced diatic endometriosis symptoms (4).

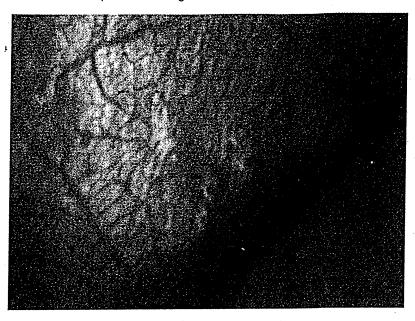
other patients underwent subsequent laparoscopic es for recurrent pelvic pain and had evaluation of hragm. Three women (cases 5, 9, and 21) had no matic endometriosis and 1 woman (case 12) had a se of superficial diaphragmatic lesions. There also ne mild, filmy adhesions between the liver and the m at the site of previous treatment.

of the patients who was not treated (case 24) later syr amatic, with excruciating left-sided chest pain er anancy after the operation. The pain worsened livery and on cessation of breast-feeding. Other or her chest pain were ruled out in a thorough work-up and it was ascribed to her diaphragmatic endometriosis. She underwent another laparoscopy, which revealed thick adhesions between the left hemidiaphragm and the dome of the liver at the site where the endometriotic lesion previously had been seen but not treated. She currently is attempting to conceive again and is using conservative pain management.

#### DISCUSSION

Several lessons can be learned from this case series. First, most patients with endometriosis involving the diaphragm have no associated symptoms. Only 7 of the 24 patients in the present series had shoulder, epigastric, chest, or right upper quadrant pain, presumably secondary to irritation of the diaphragm by endometriosis. The implants generally were superficial and caused no discomfort.

We currently cannot explain why some patients were symptomatic and some were not, especially with respect to the depth of infiltration of the lesions. Because it is not unusual for a woman to have endometriosis lesions on different areas of the pelvic cavity with no directly correlated



symptoms, the same could be true for diaphragmatic endometriosis. For example, superficial endometriosis can occur on the bowel, bladder, or ureter without intestinal or urinary symptoms.

Second, diaphragmatic endometriosis was found mostly in women with severe pelvic endometriosis. This observation may be related to the primary etiologic mechanism of this condition (8). According to the theory of retrograde menstruation, implants are expected to be found most frequently on the uterosacral ligaments and near the tubal ostium, as observed by Ishimura and Masuzaki (8). Consistent with this observation, our series showed that the presence of endometriosis on the diaphragm is a rare occurrence.

Further, endometrial implants on the diaphragm were found primarily in women with evidence of widespread disease, frequently involving the bladder, ureter, and bowel. The involvement of the appendix, ileocecum, and cecum in 41% of the patients, in addition to the presence of more dense endometrial implants in the right hemidiaphragm than the left hemidiaphragm, also is in accordance with the transportation of viable endometrial cells in peritoneal fluid following a characteristic clockwise circulation pattern, from the pelvis up the right gutter to the diaphragm, across the upper abdomen, and back down the left gutter. Although the pattern of spread of the endometriosis seems supportive of the retrograde menstruation theory (8), because the pleura arises from coelomic epithelium, the possibility that coelomic metaplasia

is responsible for the development of endometriosis on the diaphragm cannot be ruled out (9, 10).

Last, of the three patients with superficial lesions who were asymptomatic and were not treated, one became symptomatic. The possibility that the lesions may become infiltrative could be considered.

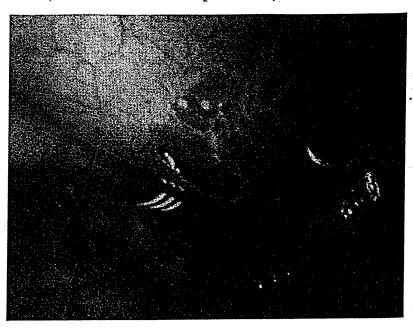
The diagnosis of diaphragmatic endometriosis before surgical exploration demands a high degree of suspicion because imaging techniques are of limited value (11, 12). Women should be asked about pleuritic, shoulder, or upper abdominal pain occurring with menses because they may not relate symptoms in these distant anatomic landmarks. Catamenial pneumothorax, a rare syndrome of recurrent pneumothoraces intimately associated with the onset of the menstrual cycle, is the most common manifestation of diaphragmatic endometriosis in cases reported in the literature (3, 12, 13), but it did not occur in any of our patients. This may stem from the fact that women presenting with catamenial pneumothorax may be referred more commonly to a cardiothoracic specialist than to a gynecologic surgeon.

One of the great advantages of videolaparoscopy is the ability to explore the diaphragm, which can be a difficult task when performing a laparotomy for the treatment of pelvic endometriosis. This benefit of videolaparoscopy always should be exploited; regardless of the indication for surgery, a complete visual inspection of the entire abdominal cavity, including the diaphragm, should be performed routinely. Examination of the abdomen occasionally results in unexpected findings. For instance, in one of the patients included

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on of an endometrial implant with the use of the CO<sub>2</sub> laser and hydrodissection.



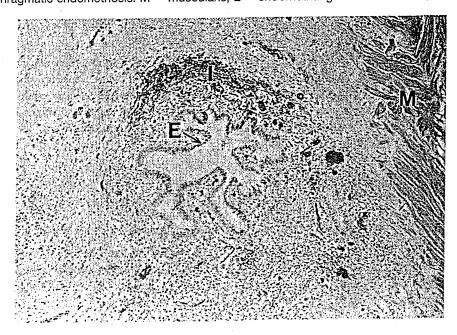
ries, a  $3 \times 4$ -cm hepatic adenoma was found on the + 'ver and was removed (case 6).

tacural history of diaphragmatic endometriosis is 1 because no prospective follow-up studies have plished. Thus, it is difficult to recommend a man-

agement strategy for this condition. When the patient is symptomatic, the need for treatment is clear. However, it remains undetermined whether asymptomatic patients should be treated to prevent the potentially severe and debilitating complications that can occur if the endometri-



gy of diaphragmatic endometriosis. M = muscularis; E = endometrial glands and stroma; I = inflammation.



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otic lesions deeply infiltrate the diaphragm. When the lesions are deep, treatment is more difficult, pain relief may not be complete, and there is a higher possibility of recurrence.

It is important to note that two of our patients who had deep and symptomatic lesions (cases 4 and 5) and who underwent conservative treatment had recurrence of their symptoms requiring subsequent treatment. Another patient (case 12) who had superficial lesions and was asymptomatic underwent second-look laparoscopy for recurrence of her pelvic pain. She was found to have a recurrence of superficial diaphragmatic lesions.

Although the symptomatic patients had acceptable pain relief and no analgesic was required for control, none had complete relief. Three patients (cases 5, 9, and 21) who were treated for diaphragmatic endometriosis (two for superficial disease and one for deeply infiltrative disease) had second-look laparoscopy for recurrent pelvic pain but did not have any recurrence of endometriosis of the diaphragm.

It generally is agreed that when treatment is deemed necessary, hormone suppression therapy should be tried first (14). However, because diaphragmatic endometriosis is an operative diagnosis, the decision to perform surgical ablation or resection of the lesion, or to defer more radical treatment and attempt medical therapy first, typically has to be made during surgery.

We recommend the surgical treatment of an incidental finding of endometriosis involving the diaphragm only if the following four criteria are met. First, the surgeon must be experienced and familiar with the treatment of deeply infiltrating endometriotic lesions. Second, the endometriotic lesions must appear to be superficial and located a safe distance from the left ventricle or the phrenic nerve. Third, support by a thoracic surgical team must be available. Fourth, the patient must have received adequate preoperative counseling regarding the risks involved in the aggressive treatment of extensive endometriosis that involves different organs.

For symptomatic patients who are unresponsive to or unable to tolerate medical suppression, and for asymptomatic patients who are concerned with the possible progress of the diaphragmatic lesions, elective treatment is preferred. This allows adequate counseling regarding the possible risks associated with the treatment.

Before surgery, the possibility of pleural or pulmonary involvement should be investigated according to the associated symptoms. The patient should be evaluated by a cardiopulmonary consultant.

In our series, the described cases were diagnosed by experienced laparoscopic surgeons in tertiary referral centers that concentrate heavily on the laparoscopic treatment of severe pelvic endometriosis, so that the risk-benefit ratio of treating the asymptomatic patients seemed to lean toward not

exposing the patients to the potentially severe consequences of endometriosis involving the diaphragm. Although it remains to be proven whether the treatment of asymptomatic endometriosis of the diaphragm is indicated, until it is proven not to be justified, we believe that when such lesions are found incidentally by surgeons who are experienced in ablating deep endometriotic lesions, and when cardiothoracic support is available, the opportunity to ablate these lesions should be considered.

Surgical management does not necessarily demand direct ablation of the diaphragmatic implants. Women with severe endometriosis who have completed their reproductive goals often prefer more radical surgery, which includes hysterectomy and bilateral salpingo-oophorectomy. However, it should be remembered that hormone replacement therapy may result in progression of the disease (6).

Three of the patients in this series underwent hysterectomy and salpingo-oophorectomy in addition to treatment of the endometriosis, and none experienced signs of recurrence.

Tubal occlusion recently was reported as a means of successfully treating spontaneous catamenial pneumothoraces; it is believed to prevent the passage of air into the peritoneum and thence into the chest (15).

The videolaparoscope is an excellent tool for treating endometriosis on the diaphragm, which is difficult to reach by laparotomy (4, 5, 7). The CO<sub>2</sub> laser is especially suitable for treating diaphragmatic endometriosis because the lesions frequently are found in places that are hard to reach, such as behind the liver. The "long arm" of the CO<sub>2</sub> laser provides unrestricted access to any lesion that is viewed through the laparoscope. Further, because CO<sub>2</sub> laser vaporization usually is achieved with good hemostasis, the need for electrocoagulation or suturing is minimized. The use of electrosurgery may be especially undesirable when the lesions are located directly below the heart, because this may induce arrhythmias or undesired heat transmission.

Another treatment modality that we used successfully in one patient is ultrasonic surgery (the CUSA). The CUSA also achieves good hemostasis while ablating the endometrial lesion, allows controlled ablation without deeply penetrating the diaphragm, and avoids electrical irritation to the heart.

When the endometriotic implant involves the entire thickness of the diaphragm, resection of the involved area with repair of any associated diaphragmatic defects is the surgical treatment of choice. When a diaphragmatic defect is formed, it is repaired with nonabsorbable sutures or staples (2).

Endotracheal intubation with controlled ventilation and a well-prepared anesthesiologist are essential because the risk that a CO<sub>2</sub> pneumothorax will develop secondary to perforation of the diaphragm is a serious concern. The operating room must be set up for the immediate insertion of a thoracic

vacuate an iatrogenic tension pneumothorax if this pecessary.

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