

Review article

Laparoscopic management of hepatic endometriosis: Report of two cases and review of the literature

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Abstract. Hepatic endometriosis is rare. Only 15 cases have been reported in the literature. All 15 were treated by laparotomy. We report two additional cases of hepatic endometriosis managed for the first time laparoscopically. Endometriosis is a progressive disease especially in women of reproductive age. One of the differential diagnoses of liver endometriosis is malignancy. Currently, there are no reports in the literature regarding complications arising from the progression of hepatic endometriosis. However, this lack of evidence does not deny its existence.

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Endometriosis is characterized by the presence of functioning endometrial tissue outside the uterine cavity. The most common locations are within the pelvis, including the ovaries, uterine ligaments, rectovaginal septum, and peritoneum.¹ Relatively uncommon, endometriosis has been described in several remote sites including the omentum, gastrointestinal tract, operative scars, lymph nodes, umbilicus, skin, lung, pleura, bladder, kidney, pancreas, and diaphragm.² The only organ in the abdominal cavity that is apparently refractory to the disease is the spleen.³

Hepatic endometriosis is rare and was first described in 1986.⁴ Fifteen cases of hepatic endometriosis have been reported in the literature.⁴⁻¹⁷ This rare condition raises several diagnostic and therapeutic challenges. When symptomatic, endometriosis of the liver is difficult to diagnose. It is often confused with other pathologies of the liver. Infrequently reported in the literature, we describe two patients with hepatic endometriosis managed laparoscopically.

Case reports

Patient No. 1

A 36-year-old nulligravid woman was referred to us because of cyclic epigastric pain for more than 1 year. Her physical examination was unremarkable without any palpable mass of the upper abdomen. Pelvic examination was unremarkable. She had no history of endometriosis. Liver function tests, blood biochemistry, and complete blood count were within normal limits. Abdominal ultrasound and computed tomography (CT) scans demonstrated a 3-cm hepatic cyst in the far caudal aspect of the right lobe of the liver. A hepatobiliary iminodiacetic acid scan of the gallbladder was normal. Pelvic sonography showed a 2-cm subserosal myoma, otherwise it was unremarkable. The patient underwent diagnostic laparoscopy for further evaluation.

During laparoscopy, extensive endometriosis involving the bladder wall, uterosacral ligaments, and pelvic peritoneum was noted. Exploration of the abdominal cavity easily revealed endometriotic implants over the diaphragm and a cystic lesion of approximately 3 cm in size on the right lobe of the liver (Figure 1A). The gallbladder appeared normal. Four trocars were placed. The operative scope with the CO₂

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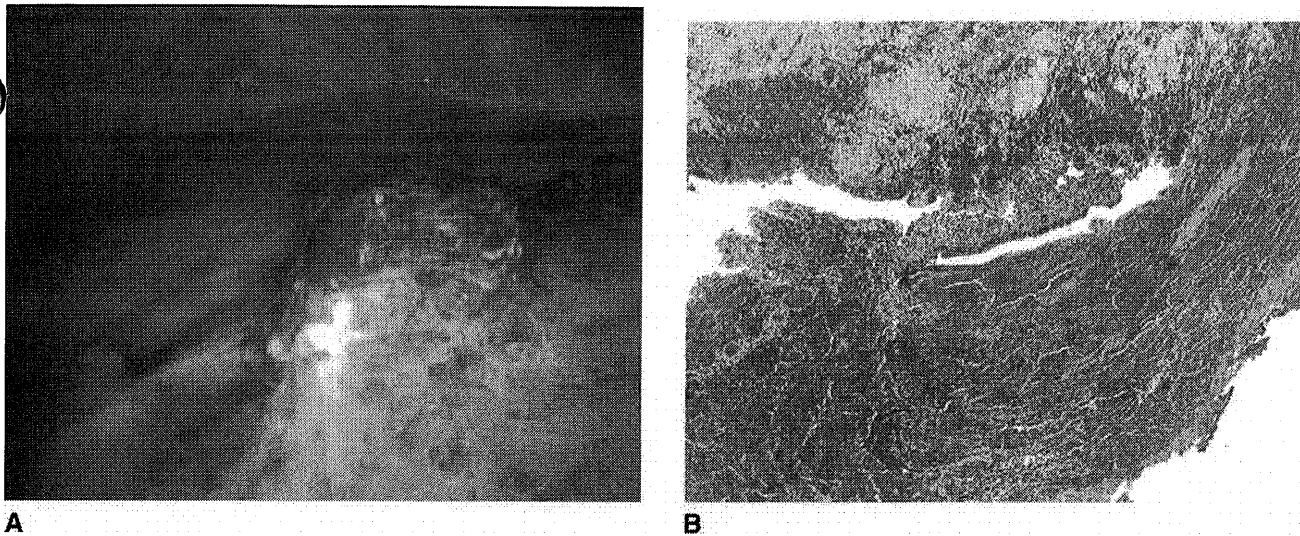


Figure 1 (A) Gross appearance of hepatic lesion. (B) Epithelial-lined glandular structure with underlying stromal elements and surrounding hemorrhage (medium-power view).

laser was placed through an 11-mm trocar intraumbilically. Additional 5-mm trocars were placed in the right and left lower quadrants. A 12-mm trocar was placed suprapubically approximately 4 to 5 cm above the pubic symphysis.¹⁸ For traction and counter traction, a grasper was used to maneuver the cyst. With the CO₂ laser set at 30 W circumferentially, the cyst was completely removed. Adequate hemostasis was acquired with the bipolar electric cautery. Frozen-section examination of the hepatic cyst was consistent with endometriosis. Following complete treatment of pelvic endometriosis, the surgery was ended. Histopathologic examination of the surgical specimen confirmed the diagnosis of endometriosis (Figure 1B). Postoperatively, the patient did well and was discharged on the same day of surgery. Immediately following surgery, she missed her menses and was found to have achieved pregnancy.

Patient No. 2

A 30-year-old nulligravid woman came to our office with symptoms of chronic pelvic pain, dysmenorrhea, and painful bowel movements. Six months prior, she had been diagnosed with stage 4 endometriosis in another center. Her physical examination and blood biochemistry were unremarkable. Preoperative abdominopelvic sonography and magnetic resonance imaging (MRI) revealed normal findings. With the presumptive diagnosis of chronic pelvic pain associated with endometriosis, endoscopic exploration was performed. Laparoscopy demonstrated severe pelvic endometriosis involving both ovaries and endometriotic implants over the intestinal serosa. The appendix was adherent to the right pelvic sidewall. Routine upper abdominal endoscopic

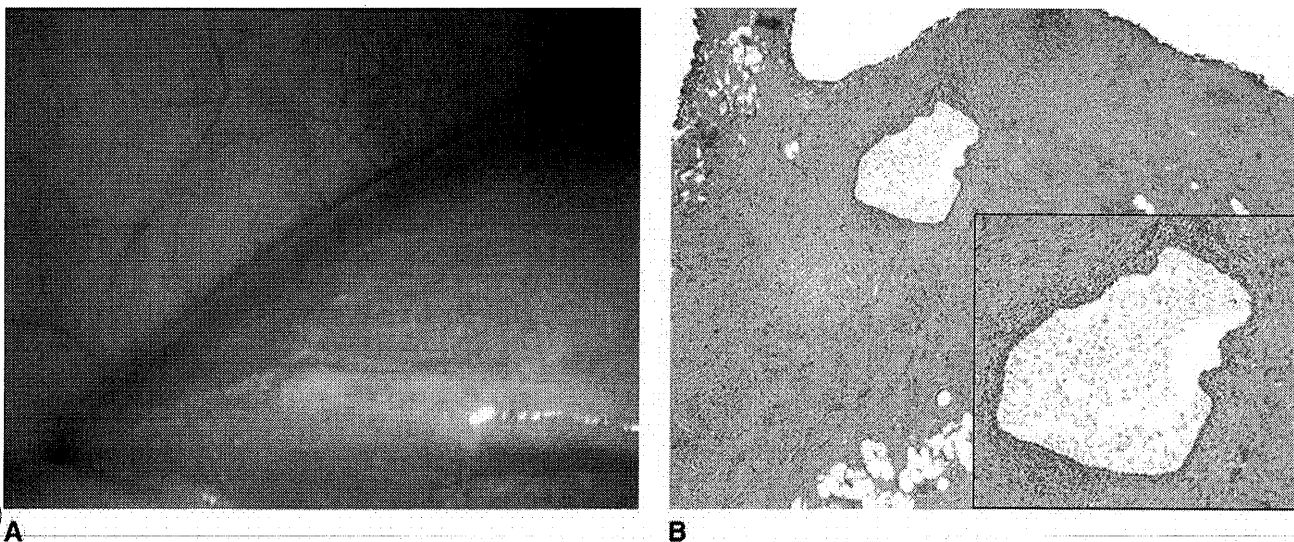


Figure 2 (A) Gross appearance of hepatic lesion. (B) A solitary glandular structure within the hepatic capsule. High-power examination reveals a focus of endometriosis with columnar epithelium and surrounding stromal cells (inset).

Table 1 Clinicopathologic features for 17 patients with hepatic endometriosis

Reference	Age (yrs)	Tumor size (cm)	Liver involvement (lobe)	Diaphragmatic involvement	HE
4	21	13	Intraparenchymal (left)	No	No
5	37	10	Intraparenchymal (left)	Yes	No
6	34	6	Extraparenchymal (right)	Yes	Yes
7	34	12	Intraparenchymal (right)	No	No
7	62	12	Intraparenchymal (left)	No	No
8	40	4	Intraparenchymal (left)	No	Yes
9	60	3	Extraparenchymal (right)	Yes	Yes
10	54	20	Incompletely intraparenchymal (right)	Yes	Yes
11	25	5	Intraparenchymal (right)	NA	Yes
12	37	3	Extraparenchymal (right)	Yes	Yes
13	31	12	Extraparenchymal (right)	NA	Yes
			Intraparenchymal (left)		
14	56	9	Intraparenchymal (left)	NA	Yes
15	52	12	Intraparenchymal (right)	No	NA
16	42	24	Intraparenchymal (right)	No	No
17	46	11	Intraparenchymal (right)	Yes	Yes
This paper, patient No. 1	36	3	Extraparenchymal (right)	Yes	Yes
This paper, patient No. 2	30	2.7	Extraparenchymal (right)	Yes	Yes

BSO = bilateral salpingo-oophorectomy; CPE = coexisting pelvic endometriosis; HE = history of endometriosis; NA = not available; RUQ = right upper quadrant; TAH = total abdominal hysterectomy.

exploration revealed a 2.7 × 2-cm solid mass over the right lobe of the liver (Figure 2A). A biopsy was taken from this lesion, and the intraoperative frozen section was consistent with endometriosis. With the same trocar placement and in the same procedural fashion as in the first patient, we laparoscopically removed the liver mass. After complete excision of the hepatic lesion, coexisting diaphragmatic endometrial implants were also treated. Pelvic endometriosis was treated as well. The patient had an uneventful postoperative recovery. Histology was reported as hepatic endometriosis (Figure 2B).

Review of the literature

Extrapelvic endometriosis is considerably less common than intrapelvic endometriosis. The occurrence decreases as the centrifugal distance from the uterus increases.⁸ Relatively common sites implicated include the gastrointestinal tract, urinary tract, and thorax.¹⁹ Hepatic location is exceedingly rare. A thorough review of the literature revealed only 15 reported cases of hepatic endometriosis.⁴⁻¹⁷ Our report adds two more cases to this rare clinical finding. Additionally, we discuss the laparoscopic approach for management of hepatic endometriosis.

Table 1 summarizes the previously reported cases and ours, comparing the clinicopathologic features. Patient age

ranged from 21 to 62 years, and lesion size ranged from 2.7 to 24 cm. The majority (94.1%) of patients were symptomatic, generally with right upper quadrant (RUQ) abdominal pain. Only in one patient was a hepatic mass incidentally discovered during follow-up sonogram for pelvic endometriosis. Ten of 17 patients had a history of endometriosis. The symptoms and preoperative findings did not suggest endometriosis in 13 patients. In four patients, percutaneous biopsy of the hepatic mass revealed endometrial tissue. Except for one, all patients reported on in the literature underwent laparotomy for the diagnosis and treatment of hepatic endometriosis. In one case, the patient refused surgery and was treated with danazol.¹¹ Including our patients, only five were operated on with the size of the lesion less than 5 cm. This reveals that early diagnosis may require a high degree of suspicion. Including our patients, associated pelvic endometriosis was reported in six of nine patients with liver endometriosis. Pelvic cavities were not evaluated in the remaining eight patients. Additionally, together with the two patients reported on here, there were evidences of diaphragmatic endometrial implants in eight patients.

Discussion

Many theories including the implantation theory, coelomic metaplasia theory, induction theory, and autoimmune theory

Symptoms and signs	CPE	Previous pelvic operation	Pathology	Treatment
RUQ pain	NA	Fallopian tube cyst removal	Hepatic endometrioma	Cyst enucleation by laparotomy
Painful RUQ mass	Yes	No	Hepatic endometrioma	Left lateral segmentectomy by laparotomy
Cyclic RUQ pain	No	No	Hepatic endometriosis	Tumorectomy by laparotomy
Acute abdomen	NA	No	Hepatic endometrioma	Hemihepatectomy by laparotomy
RUQ pain	NA	No	Hepatic endometrioma	Segmental liver resection by laparotomy
Asymptomatic	Yes	Left ovarian cystectomy due to endometriosis	Hepatic endometrioma	Cyst enucleation by laparotomy
RUQ pain	No	TAH, BSO	Adenosquamous carcinoma arising in hepatic endometriosis	Segmental liver resection by laparotomy
RUQ pain	No	TAH, BSO	Adenosarcoma arising in hepatic endometriosis	Segmental liver resection by laparotomy
Pelvic pain, rectal hemorrhage	Yes	Treatment of pelvic endometriosis	Hepatic endometriosis	Danazol therapy
Upper abdominal pain	NA	TAH, BSO	Hepatic endometrioma	Segmental liver resection by laparotomy
Noncyclic abdominal pain	NA	TAH, BSO	Hepatic endometrioma	Removal of cyst by laparotomy
Epigastric pain	NA	TAH, BSO	Hepatic endometrioma	Left hepatic lobectomy by laparotomy
RUQ pain	NA	TAH, BSO	Hepatic endometrioma	Right hemihepatectomy by laparotomy
Acyclic Epigastric pain	No	No	Hepatic endometrioma	Removal of cyst by laparotomy
RUQ pain	NA	Endometriosis surgery, subtotal hysterectomy	Hepatic endometrioma showing moderately atypical complex hyperplasia	Right hemi-hepatectomy by laparotomy
Cyclic RUQ pain	Yes	No	Hepatic endometriosis	Cyst enucleation by laparoscopy
Chronic pelvic pain	Yes	Treatment of pelvic endometriosis	Hepatic endometriosis	Laparoscopic excision

have been proposed for the pathogenesis of endometriosis.^{7,16,20} It is likely that a combination of these theories may apply to each individual patient. Hepatic endometriosis was found more often in patients with severe pelvic endometriosis in those whose pelvic cavity was evaluated. This was also valid in our cases. This observation may be related to the primary etiologic mechanism of this condition. Additionally, the presence of endometrial implants more on the right hepatic lobe than the left lobe is also 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 endometriosis seems supportive of the retrograde menstruation theory, the possibility that coelomic metaplasia is responsible for the development of endometriosis on the liver cannot be ruled out. This might indicate that the peritoneum covering the surface of the liver achieves its metaplastic potential and subsequently proliferates into the liver parenchyma. These two theories fail to explain why in some cases hepatic endometrioma is solely intraparenchymal. We believe that lymphovascular spread also may play an important role. Supporting this, most of the patients in the reported cases had previously undergone pelvic surgery for endometriosis, as seen in Table 1. Thus, lymphovascular spread of endometrial fragments offers a better explanation than the other theories for the patients

having complete intraparenchymal endometriosis of liver. As to its etiology, there is much more to learn from endometriosis occurring in unusual sites and instances.

The diagnosis of extrapelvic endometriosis is difficult, and it is often made many years after the onset of symptoms. On the other hand, hepatic endometriosis is so uncommon that such a diagnosis before surgical exploration demands a high degree of suspicion. Women should be evaluated for upper abdominal pain associated with the onset of the menstrual cycle. However, this is not the most common manifestation of the hepatic endometriosis encountered in the cases reported in the literature. Excluding one, all patients described in the literature had epigastric or RUQ pain; and, including one of our patients, only two patients complained of characteristic cyclic pain related with menses (Table 1). These findings demonstrate that medical history regarding pain may not be that helpful in diagnosing hepatic endometriosis. Although ultrasound, CT, and MRI aid in the diagnosis, the final diagnosis can be made only by pathologic evaluation. Only 5 out of 15 cases reported in the literature had correct preoperative diagnosis. The presumptive diagnoses were most often echinococcal or amebic cyst, pyogenic abscess, cystadenoma, hematoma, or metastatic disease. Percutaneous biopsy of the hepatic mass revealed endometrial tissue in four patients. In one patient with cyclic abdominal pain, a presumptive diagnosis of endometriosis was made based on history and imaging findings alone.⁶

Because diagnostic laparoscopy had been planned, none of our patients had percutaneous biopsies, and the diagnosis was made during the laparoscopic surgical intervention.

There are no other reported cases of laparoscopically treated hepatic endometriosis in the literature. One of the significant advantages of laparoscopy is the ability to explore the upper abdominal region, which can be a difficult task when performing a laparotomy for the treatment of pelvic endometriosis.²¹ This benefit of laparoscopy always should be exploited. Regardless of the indication for surgery, complete visual inspection of the entire abdominal cavity, including the upper abdominal region, should be performed routinely. Furthermore, by this report we also demonstrated that laparoscopy could be a reliable treatment modality for extraparenchymal hepatic endometriosis. Like other minimally invasive procedures, laparoscopic management of hepatic endometriosis has potential benefits of less postoperative pain, a shorter hospital stay, earlier mobilization, and a quicker recovery period. Regarding our cases, laparoscopic approach was successful with no conversion to open surgery. We believe that local excision may be an appropriate choice for hepatic endometriosis.

The natural history of hepatic endometriosis is unknown because no prospective follow-up studies have been published. In patients with superficial lesions who are asymptomatic and are not treated, one should consider the possibility that these lesions may become infiltrative and symptomatic. Thus, it is difficult to recommend a management strategy for this group of patients. 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 endometriotic lesions deeply infiltrate the liver. Furthermore, although both gonadal and extragonadal endometriosis are generally benign conditions, malignancy occurring within endometriotic foci has been well documented.²² One study described a patient with endometrioid adenosquamous carcinoma,⁹ and another study reported a case of adenocarcinoma arising in hepatic endometriosis.¹⁰ Recently, researchers reported a hepatic endometrioma showing moderately atypical complex hyperplasia.¹⁷ Because extrapelvic disease may also represent a metastatic spread, one must always be aware that in unusual involvement sites malignancy must be excluded. A pathology consultation in the operating room would be very helpful in this regard.

Conclusion

This report demonstrates the importance of not ignoring the thorough exploration of the abdominal cavity in patients undergoing laparoscopic pelvic surgery. Also for upper abdominal surgeons, it is important not to ignore the rest of the abdomen and pelvis, especially in patients with endometriosis. Although rarely seen, hepatic endometriosis can be accurately diagnosed and appropriately treated if the surgeon has an increased awareness and high index of suspi-

cion during laparoscopy. The laparoscopic approach may be recommended as an option for the treatment of hepatic endometriosis in proper settings. Currently, there are no reports in the literature regarding the progression of hepatic endometriosis, but the lack of its evidence does not deny its existence. As time goes by, we might encounter more evidence of liver involvement and subsequently more severe symptoms, which we are hoping to prevent with our recommendation for its treatment.

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