

Giant endometrial cyst of the liver: a case report and review of the literature

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SUMMARY: Giant endometrial cyst of the liver: a case report and review of the literature.

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Endometriosis is a benign condition described as the presence of endometrial-like tissue found outside the uterine cavity. Hepatic endometriosis is one of the rarest localizations of extrapelvic endometriosis, only 22 cases have been reported in the literature. The preoperative

diagnosis of hepatic endometriosis is rather difficult because in about the half of the patient affected they had no history of endometriosis. Moreover radiological images reveal no characteristic findings for hepatic endometriosis. It is often described as cystic mass with or without solid component, difficult to distinguish from hepatic abscess, hematoma, cystadenoma or malignant neoplasia. We report a case of a 27-year-old female with a large cystic mass involving the left lobe of the liver. The patient underwent laparoscopic exploration and converted to laparotomy for resection of giant hepatic endometriosis.

KEY WORDS: Endometriosis - Liver cyst - Hepatic endometriosis.

Introduction

Endometriosis is characterized by the presence of functioning endometrial tissue outside the uterine cavity (1). The most common locations are within the pelvis, including the ovaries, uterine ligaments, rectovaginal septum, and peritoneum. 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 (2-3). The only organ in the abdominal cavity that is apparently refractory to the disease is the spleen. Hepatic endometriosis is rare and was first described in 1986 (4). To date 22 cases of hepatic endometriosis have been reported in the literature (5-21). 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. We describe the case of a patient with giant hepatic endometriosis.

Case presentation

A 27-year-old nulligravida woman was referred to us because of abdominal mass that patient discovered 3 months before, without abdominal pain associated with the onset of the menstrual cycle, but constant tenderness on upper and low left abdominal quadrants (Figure 1). Physical examination demonstrated liver function tests, blood biochemistry, complete blood count, serology for echinococcal disease and the tumors markers were within normal limits. She had no history of endometriosis. Preoperative abdominal ultrasound, computed tomography scan (CT) and magnetic resonance imaging (MRI) demonstrated a 30-cm hepatic cyst in the left lobe of the liver that reached segments IV, V and VIII (Figure 2). The patient underwent diagnostic laparoscopy that excluded other peritoneal malignant or benign tissue deposits. Surgical exploration highlighted a giant cystic neoplasia, originated from left liver, that caused atrophy and shifted on left liver hepatic parenchyma (Figure 3), reached segments V and VIII and beneath covered the hepatoduodenal peduncle. Laparoscopy was converted in open approach and median laparotomy was performed; Pringle manoeuvre was prepared but not executed. Complete mobilisation of left liver and partial of right were performed. Atypical left hepatectomy tailored on right margins of lesion was performed. Va-

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Fig. 1 - Abdominal swelling is more evident in epigastric region and left side of the abdomen.



Fig. 2 - Computerized Tomography showing a voluminous cystic mass of the left lobe of the liver involving also the segments IV and V.

scular and biliary stricture were secured by intrahepatic glissonian approach; left hepatic vein by transparenchymal approach. Histopathologic examination and immunostaining of the surgical specimen established the diagnosis of endometriosis. Postoperatively, the patient did well and was discharged on seventh day of surgery. Immediately following surgery, she missed her menses and became pregnant.

Discussion

Extrapelvic endometriosis is significantly less common than intrapelvic endometriosis. It occurs more frequently in women of reproductive age (10%) than in postmenopausal women (2,5%) (22). Unusual sites include the gastrointestinal tract, kidney, bladder, lungs, heart, pleura (23). Hepatic location is rare. A review of

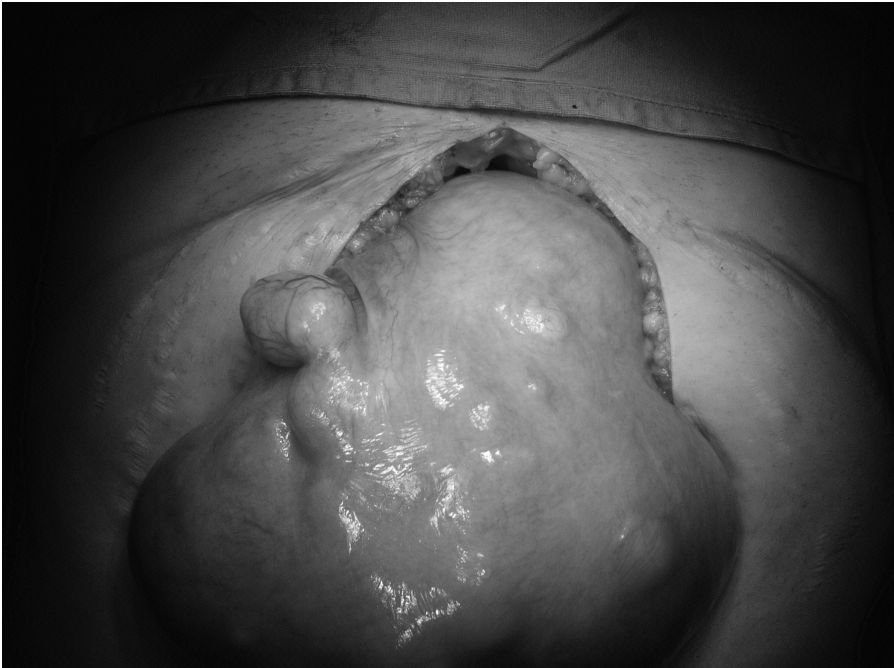


Fig. 3 - Gross appearance of hepatic lesion.

the literature revealed only 21 (4-21) cases of hepatic endometriosis (Table 1). Mean age was 39 years and ranged between 21 and 62 years, lesion size ranged from 2.7 to 30 cm. Our report add one more rare case with a lesion sized 30 cm on diameter. The number of cases in which there was involvement of the left liver was almost equal to that of the right (11 vs. 10). One case presented with bilobar involvement. Nine patients had a history of endometriosis. Surgeons included endometriosis in the process of differential diagnosis only for four patients. Laparotomy was useful for the diagnosis of endometriosis in all patients with the exception of four cases, where a liver biopsy was performed. The pathogenesis of hepatic endometriosis is unclear; considering all the theories proposed each satisfies only some of the reported cases. The case we present about a large lesion located into the hepatic parenchyma could be better explained by lymphovascular spread of endometriotic cells. This theory, as demonstrated by presence of endometrioid cells in lymphatic vessels or locoregional lymph nodes in patients with infiltrating endometriosis, could explain intraparenchymal cases of liver endometriosis (26). Indeed no relation was found between the cyst and peritoneal surface, refusing the coelomic metaplasia theory that indicate the potentiality of microenvironment of the peritoneum to change connective tissue to endometrial tissue. The absence of previous history of endometriosis for our patient makes the implantation theory unlikely. Extrapelvic endometriosis is rare and its diagnosis is often made many years after the onset of symptoms, because only few patients presented

characteristic cyclic pain related with menses. Although ultrasound, CT and MRI are helpful, no typical image of endometriosis cyst has been described so the final diagnosis can be made only by histological evaluation (27). The working diagnosis were both benign conditions, as echinococcal cyst, abscess, hematoma, cystadenoma, and malignant cystic neoplasm, as cystadenocarcinoma or metastatic disease. In our case we carried out a laparoscopic exploration. The additional benefit to notorious advantages of laparoscopic was the possibility of identifying other abdominal tissue deposits and, in case of endometrioid seeding, of treating those by laparoscopy or of avoiding a useless laparotomy in case of malignant carcinosis. Abnormal size of lesion led us to a conversion in open approach for surgical treatment of hepatic lesion. Resection of cystic endometriosis should be always considered in symptomatic patients. However the management strategy of endometriosis of the liver is so controversial because it's unknown the natural history of hepatic endometriosis due the lack of prospective study. Malignant transformation of endometriosis is a rare event, occurring commonly in the ovary; however two cases of sarcomas and one case of adenocarcinoma arising from endometriosis of the liver have been described (28-30).

Conclusion

The exploration of the abdominal and pelvic cavity should be the considered the first step helpful for sug-

TABLE 1 - CLINICAL AND PATHOLOGICAL FEATURES OF CASE REPORTS OF HEPATIC ENDOMETRIOSIS.

Reference	Age (years)	Liver involvement	Endometrial history	Maximum size (cm)	Preoperative diagnosis
Finkel et al.	21	Left lobe	No	13	No
Grabb et al.	21	Left lobe	No	NA	No
Rovati et al.	37	Left lobe	Yes	NA	Yes
Verbeke et al.	34	Right lobe	No	12	No
Verbeke et al.	62	Left lobe	No	12x10x7,5	No
Chung et al.	40	Left lobe	Yes	6,4x3x2,5	No
Inal et al.	25	Right lobe	Yes	5	No
Huang et al.	56	Left lobe	Yes	9x6	No
Khan et al.	31	Bilobar	Yes	NA	Yes
Khan et al.	59	Right lobe	Yes	NA	Yes
Tuech et al.	42	Right lobe	No	22x24x30	No
Jelovsek et al.	52	Right lobe	Yes	11x7	Yes
Fichet et al.	45	Right lobe	No	17x13x15	No
Nezhat et al.	36	Right lobe	Yes	3	No
Nezhat et al.	30	Right lobe	Yes	2,7x2	No
Goldsmith et al.	48	Left lobe	Yes	11x13	No
Schuld et al.	39	Right lobe	No	6,8x2,3	No
Fluegen et al.	32	Right lobe	No	9,5x12	No
Rivkine et al.	51	Left lobe	No	8x4,5x4	No
Bourasa et al.	35	Left lobe	No	10	No
Liu et al.	36	Left lobe	No	6,5x6	No
This paper	27	Left lobe	No	30	No

gesting intervention in cases of hepatic lesion of uncertain origin, especially in patients with history of endometriosis. Due to the difficult interpretation of radiographic images and the atypical clinic, diagnosis of hepatic endometriosis is entrusted to the skill and the high degree of suspicion of the surgeon.

Competing interest

The authors have no conflicts of interests.

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Author's contribution

MD, FD and AI participated in the surgery of this case and AI proposed to describe the case. MD wrote the manuscript and provided the pathological images. FD and GM searched previous reports and produced the review table. All authors read and approved the final manuscript.

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