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Case Report

Subcapsular hepatic endometriosis: case report and review of the literature

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ABSTRACT

Hepatic endometriosis is a very rare medical condition characterized by the implantation of ectopic endometrial tissue within the hepatic parenchyma. Preoperative diagnosis is difficult via cross-sectional imaging and histopathologic evaluation remains the gold standard for diagnosis. We report a case of hepatic endometrioma in a 44-year-old woman with history of endometriosis. The literature is reviewed, and magnetic resonance imaging findings together with differential diagnosis of hepatic endometriosis are highlighted.

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Case report

A 44-year-old woman with a history of endometriosis was referred to our radiology unit with progressive right upper quadrant pain and vomiting for few months. The pain is periodic and dull in nature. Her surgical history included hysterectomy for endometriosis and cholecystectomy. The patient had a previous ultrasound that reported a 3-cm complex cystic lesion in the right lobe of the liver which was not further characterized.

Her tumor markers (alfa fetoprotein, CA 19-9, CA 125 and carcinoembryonic antigen) and liver function tests were within normal range.

Computed tomography (CT) scan of the abdomen demonstrated 3 cm rather well-defined hypodense subcapsular lesion in the right lobe of the liver that illustrated heterogeneous peripheral enhancement in the venous and delayed phases (Fig. 1). The primary differential considerations were subcapsular abscess, granuloma, hematoma, or metastasis, further characterization with magnetic resonance imaging (MRI) was suggested.

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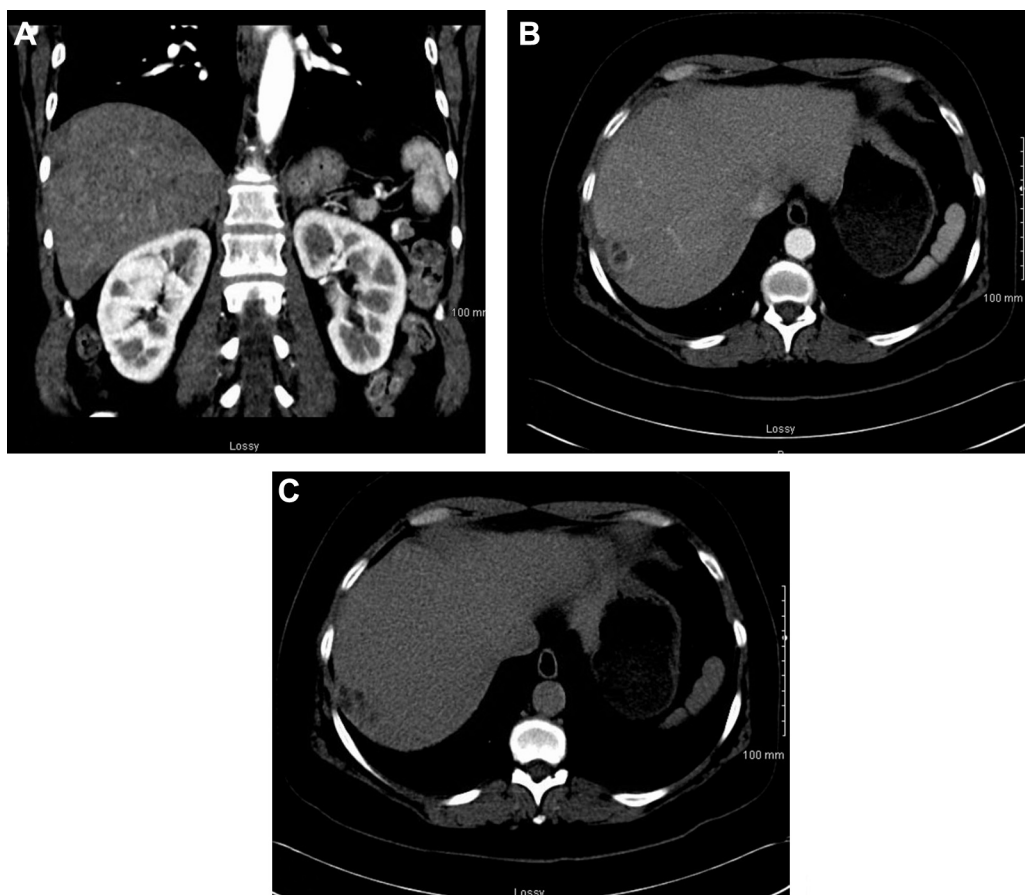


Fig. 1 – A 44-year-old woman with periodic right upper quadrant pain. Technique: multiphasic CT scan of the liver after injection of 90-mL Omnipaque 350 IV contrast obtained in arterial phase (after 25 seconds) coronal images (A), venous phase axial images (after 60 seconds; B) and delayed phase axial images (after 3 minutes; C). Findings: rather well-defined subcapsular hypodense focal lesion demonstrates faint arterial enhancement (A) with heterogeneous peripheral enhancement in venous (B) and delayed phase (C). There is no complete filling in delayed phase. No calcification. No washout.

MRI examination revealed subcapsular partially cystic focal lesion with intrinsic high-signal intensity in both T1-weighted and T2-weighted images—with and without fat suppression—suggestive of subacute hemorrhagic content. It exhibits heterogeneous peripheral enhancement in multiphasic contrast study (Fig. 2). The differential diagnosis was hematoma, complex hepatic cyst, hepatic adenoma (probably due to blood products), and hemorrhagic metastasis (ie, melanoma).

Due to nonconclusive imaging findings, CT-guided core biopsy was performed. Histopathology reported endometrioid glands, stroma, and smooth muscle, consistent with adenomyoma. The patient had followed up for 2 years without considerable increase in the size of the endometrioma, but with persistent pain in spite of hormonal treatment, she finally underwent hepatic segmentectomy for segment VII, and histopathology report confirmed the diagnosis of hepatic endometrioma (Fig. 3).

Discussion

Endometriosis is a common benign disease affecting women of reproductive age usually with an estimated prevalence

rate of 17%-47% among infertile women [8]. Uterine and extrauterine endometriosis was the first described by Rokitansky in 1860 [9]. Endometriosis is usually confined to the pelvis and reproductive organs, the ovaries are the most frequent location; however, other remote sites including the gastrointestinal tract, peritoneum, chest, scar tissue, lymph nodes, and kidneys have been also described [3–6,8]. The clinical presentation of the disease is variable and can be associated with distressing symptoms such as pelvic pain, dyspareunia, infertility, or it may be asymptomatic and incidentally discovered [8].

The mechanism of extrauterine endometriosis is still uncertain [7,8]. However, various theories have been proposed to explain the pathogenesis of endometriosis. In our report, we are discussing 2 of the major theories that strengthen their hypothesis through providing strong supporting evidences. These theories are the implantation theory and the celomic metaplasia theory.

The implantation theory suggests that endometrial tissue is transplanted into the peritoneum and pelvic organs through retrograde menstruation, hematogenous and/or lymphatic dissemination, or iatrogenic injury [7,8]. Considerable evidences have validated this theory: (1) The menstrual effluent

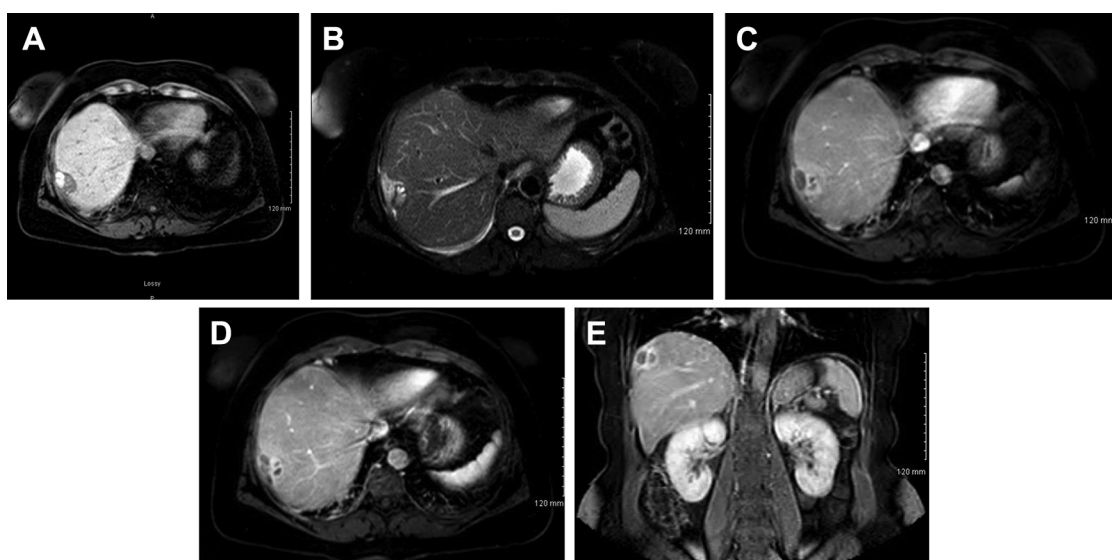


Fig. 2 – A 44-year-old woman with periodic right upper quadrant pain. Technique: Multisequence Multiplanar MRI study of the liver before and after administration of 18-mL Multihance IV contrast. Findings: rather well-defined subcapsular complex focal lesion illustrates hyperintense signal in T1WI with fat saturation axial images (A) and hyperintense signal in T2WI with fat saturation axial images (B). Postcontrast injection demonstrates peripheral heterogeneous enhancement in arterial phase (after 15 seconds) axial images (C), venous phase (50 seconds) axial images (D), and delayed phase (after 3 minutes) coronal images (E).

and peritoneal fluid usually have viable endometrial cells, (2) endometrium can experimentally be implanted and grown in a peritoneal cavity, and (3) adequate percentage of female have a degree of retrograde menstruation.

In celomic metaplasia theory, few authors considered peritoneal endometriosis, endometriosis of the ovary and endometriosis of rectovaginal septum as 3 separated entities each has a different pathogenesis [7,8]. The supporting evidence of this theory proposed that the peritoneal endometriosis originates from the metaplasia of peritoneal mesothelium, ovarian endometriosis results from either invagination of ovarian cortex, or metaplasia of celomic epithelium and the rectovaginal nodule from metaplasia of Müllerian duct.

Hepatic endometriosis, first described by Finkel et al. in 1986, is a rare entity of extra-uterine endometriosis [1–3]. Within the reviewed literature, only 21 cases were reported [4].

Although ovarian endometriosis usually illustrates classical radiological findings, there is no specific diagnostic sign to distinguish hepatic endometriosis from other hepatic lesions [4]. Accordingly, the histopathologic examination is considered the gold standard for definite diagnosis.

Within the reviewed literature, variable differential diagnosis of hepatic endometriosis was provided depending on clinical presentation and the radiological characteristics of the lesion, for eg, hematoma, complex cyst, metastasis, and hepatocellular carcinoma. Interestingly, they all described it primarily as a complex cystic lesion on ultrasound with distinctive subcapsular location in subsequent imaging. In our case, the lesion was complex cyst in a reported ultrasound and subcapsular in location on CT and MRI images.

We summarized the published cases and their respective described findings in (Table 1) below.

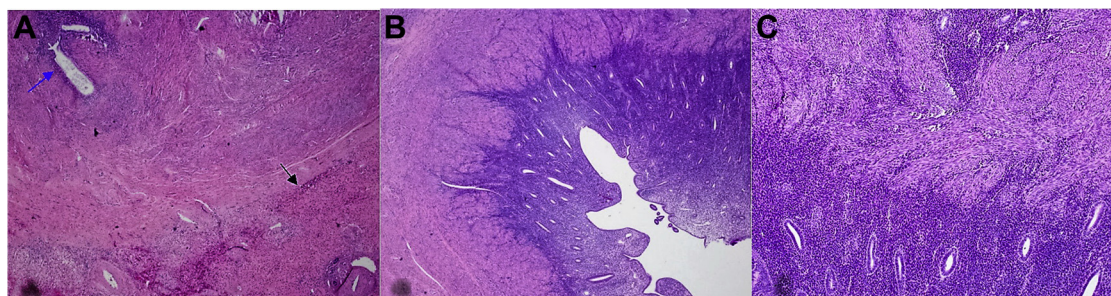


Fig. 3 – Histopathology findings: (A) adenomyoma (blue arrow) and adjacent liver parenchyma (black arrow; H&E, 40 $\times$). (B) Adenomyoma in liver with endometrioid glands, stroma, and smooth muscle tissue (H&E, 100 $\times$). (C) Higher power image of the adenomyoma showing endometrioid glands, stroma, and smooth muscle tissue (H&E, 200 $\times$).

Table 1 – Features of reported cases of hepatic endometriosis in literature.

Author/reference	Age (year)	Symptom and sign	Pain related to menstruation	Location of the mass	Coexisting endometriosis	Previous pelvic operation	Treatment
Finkel/[2]	21	Epigastric and RUQ mass	N/A	Subcapsular left lobe hepatic cyst	None	Removal of fallopian tube cyst	Cyst enucleation
Grabb/[12]	21	Chronic epigastric pain with nausea and vomiting hepatomegaly with right subcostal mass	N/A	Subcapsular left lobe hepatic cyst	None	Fallopian tube cyst removal 3 years before	Deroofing, danazol
Rovati/[9]	37	Chronic epigastric pain, epigastric mass	N/A	Subcapsular left lobe hepatic cyst	Left ovary, peritoneum	None	Left lateral segmentectomy, danazol
Verbeke/[10]	34	Acute abdomen	N/A	Subcapsular right lobe hepatic cyst	None	None	Excision
Verbeke/[10]	62	Right epigastric pain	N/A	Subcapsular left lobe hepatic cyst	None	Abdominal operation for Meckel's diverticulum in early childhood	Excision
Cravello/[13]	34	Cyclical pain	N/A	Subcapsular right lobe hepatic cyst	Yes	None	Metastectomy
Weinfeld/[11]	60	Right upper abdominal tenderness	N/A	Right lobe, falciform ligament	Both ovaries, pouch of Douglas	Hysterectomy and bilateral oophorectomy 23 years before, resection of endometriosis adjacent to the urinary bladder 4 y before	Excision of right lobe tumor, left hepatectomy
Chung/[14]	40	Asymptomatic	N/A	Subcapsular left lobe hepatic cyst	yes	Ovarian cystectomy	Segmentectomy
Inal/[15]	25	Pelvic pain	N/A	Subcapsular right lobe hepatic cyst	Pelvic endometriosis	None	Danazol
N'senda/[18]	54	Abdominal pain	N/A	Subcapsular right lobe hepatic cyst	No	N/A	Right hepatectomy
Jeanes/[19]	31	Abdominal pain	N/A	Bilobar	Yes	Yes	Right hepatectomy
Khan/[16]	31	Malaise, jaundice, abdominal distension	N/A	Bilobar	Yes	Hysterectomy and bilateral oophorectomy	En bloc removal of right lobe mas, left lobe mass left
Khan/[16]	59	RUQ pain + hepatomegaly	N/A	Subcapsular right lobe complex hepatic cyst	yes	Removal of ruptured cyst	Right hepatectomy
Haung/[3]	56	Epigastric pain + tender RUQ mass	No	Subcapsular left lobe complex hepatic cyst	Bilateral ovaries, uterine cervix and Pouch of Douglas	Hysterectomy and bilateral oophorectomy	Left hepatectomy
Tuech/[20]	42	Asymptomatic	No	Subcapsular right lobe	No	N/A	Cyst excision
Reid/[21]	46	N/A	N/A	Subcapsular right lobe hepatic cyst	Yes	N/A	Right hepatectomy + goserelin
Groves/[1]	52	RUQ pain	N/A	Right hepatic lobe	N/A	Hysterectomy/oophorectomy	Right hemihepatectomy

Goldsmith/[6]	48	RUQ pain	No	Subcapsular complex cyst segment IV and VIII (right lobe)	Yes	Hysterectomy and bilateral salpingo-oophorectomy	Nonanatomical resection using Cavitron ultrasonic aspirator
Asran/[5]	61	Epigastric Pain	N/A	Scattered throughout liver	Yes	Hysterectomy/right salpingo-oophorectomy/bowel loops resection	N/A
Schuld/[22]	39	Bronchobiliary fistula	No	Subcapsular right lobe	No	No	Segmentectomy
Fluegen/[23]	32	RUQ pain	No	Subcapsular right lobe	No	No	Pericystectomy
Rivkine/[23]	51	Epigastric pain and vomiting	No	Subcapsular left lobe	No	Hysterectomy for leiomyoma	Left hepatectomy and diaphragm resection
Pallarés/[17]	41	Right hypochondrial pain	Yes	Right lobe	N/A	N/A	Laparoscopic surgical removal
Kai et.al/[4]	36	RUQ pain	Yes	Left lobe, subcapsular segment III	No	No	Exploratory operation + pericystectomy
Incomplete/2016 current report	35	RUQ pain	Yes	Subcapsular complex cyst segment VII. Right lobe	Yes, history of endometriosis	Hysterectomy	Hepatic segmentectomy

Conclusions

The radiologist should consider hepatic endometrioma on the top of the differential diagnosis of complex hepatic cyst, subcapsular in location with hemorrhagic content in any women, particularly of reproductive age with prior history of endometriosis.

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