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Case Report General Surgery

Is it resistant? A case report of splenic endometriosis and review of the literature

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ABSTRACT

Endometriosis is an estrogen-dependent benign disease that generally affects women at childbearing age. Despite its high incidence, pathogenesis of endometriosis is poorly clarified. There are several theories explaining the mechanism of disease such as retrograde menstruation, coelomic metaplasia, vascular and lymphatic metastasis. While endometriosis has been described in many intra- and extra-abdominal organs including the ovaries, omentum, colon, liver, lungs and brain, parenchymal splenic endometriosis is a rare situation. In recent years, with reported cases, this rare condition came onto the scene. We also wanted to present our case and question the idea of splenic immunity against endometriosis according to current literature. From now on, splenic endometriosis can be thought of as a differential diagnosis for unidentified lesions of spleen.

Keywords: Endometriosis; laparoscopy; spleen; splenectomy.

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Endometriosis is a common gynecological disorder defined as the presence of endometrial tissue outside the uterine cavity. The prevalence of endometriosis among reproductive women is 6-10% [1]. Nevertheless, due to unspecified symptoms and need for direct visualization and pathologic examination of the implant, endometriosis is an underdiagnosed disease [2].

Despite its high incidence, pathogenesis of endometriosis is poorly clarified. Retrograde menstruation, the most accepted theory, suggests backflow of endometrial lining and implantation of the viable endometrial tissue in pelvic organs [3]. However, extra-pelvic implantations failed to be explained by this theory. Additionally, prevalence of endometriosis remains very low regarding prevalence of retrograde menstruation and this data supports the multifactorial basis of the disease including genetic and environmental features [3]. Other theories include coelomic metaplasia and vascular/lymphatic metastasis, which are more coherent regarding extra-pelvic implantations [3]. In addition, iatrogenic endometriosis can be seen especially in women after gynecologic operations such as myomectomy, hysterectomy, and cesarean section [4].

A multicentric prospective study including premenopausal women undergoing laparoscopy without a previous surgical diagnosis of endometriosis showed that the incidence of endometriosis ranges between 35% and 100% in symptomatic women [5]. Common implanta-

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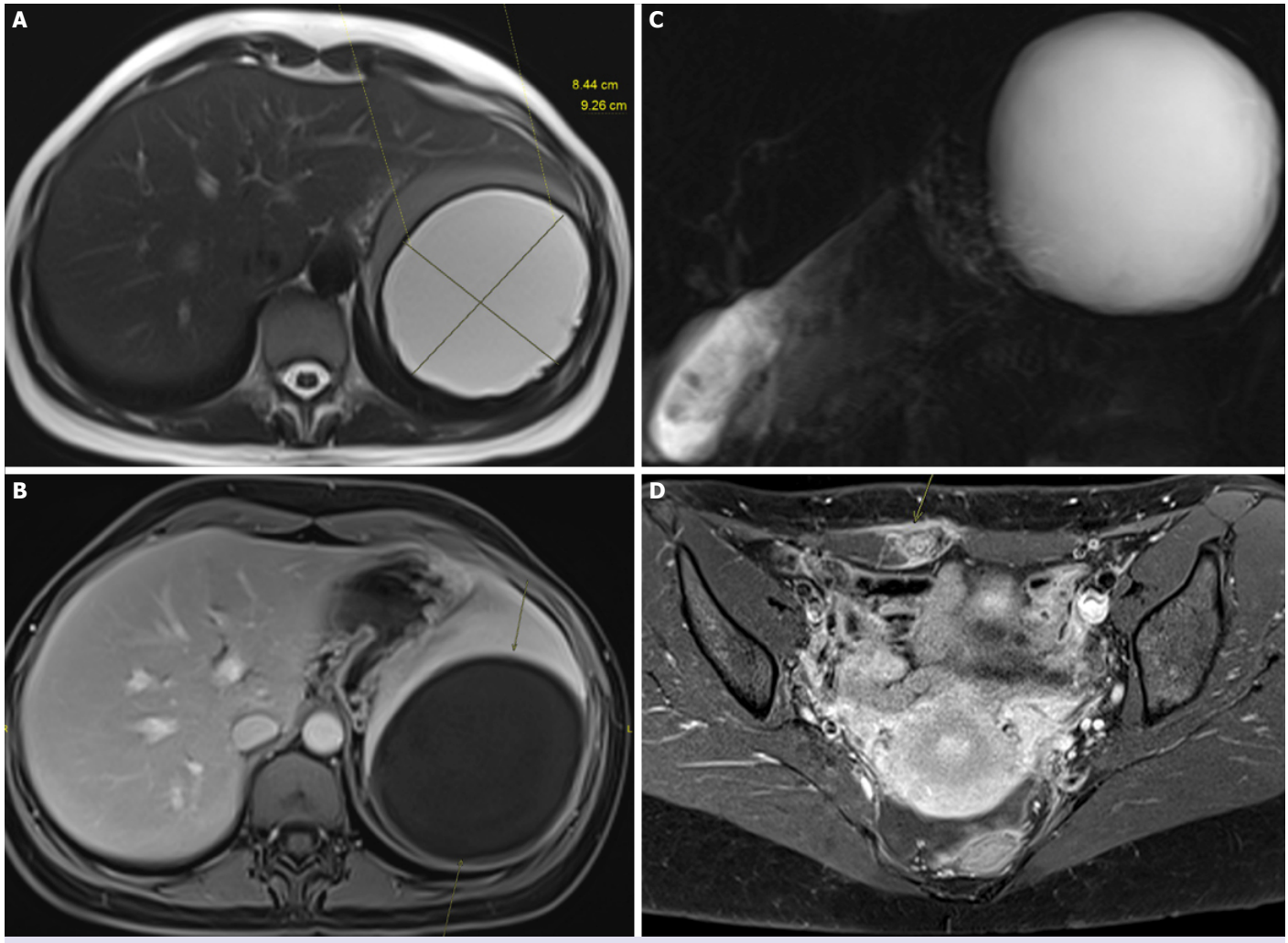

Figure 1. MRI: An 8.44×9.26 cm lesion with sharp borders within the spleen parenchyma which appears hypointense in axial T1 sequence **(A)** and hyperintense in coronal T2 sequence **(B)**. T2 sequence revealed minimal border irregularity on the inner side of the cyst. MRI cystographic investigation revealed a cystic lesion in the spleen parenchyme **(C)**. Post-contrast axial fat suppressed T1 investigation revealed a heterogenous lesion medial to the right rectus muscle **(D)**.

tion sites for endometrial tissue are pelvic structures such as the ovaries, ligaments of the uterus and peritoneum. Rarely, endometrial tissue may spread to distant organs beyond pelvis including the omentum, skin, vertebrae, lungs, liver, skeletal muscle and even to brain [2]. Exceptionally, spleen is the only organ mentioned as refractory to development of endometriosis [6, 7].

Contrary to the idea of splenic immunity against endometriosis, three cases of splenic endometriosis have been reported since 2020 [8–10]. Interestingly, all the reports claim to be the first report of the literature. With this case report and review of the literature, we wanted to question the idea of immunity of spleen against endometriosis and create a source for further research.

CASE REPORT

A 31-year-old primigravida female patient presented with swelling and intermittent abdominal pain in the right lower quadrant for two months. She had a history of solid mass excision via cesarean section due to 22x15 cm mucinous cystadenoma in her right ovary. On physical examination, the right lower quadrant was tender, and a palpable mass was found in the right lower quadrant at the upper part of the Pfannenstiel incision. Ultrasonography was planned for further evaluation, and preoperative ultrasonography demonstrated a hypoechoic lesion 22x38 mm in size at the level of right rectus muscle and a cystic mass of 90x85 mm in size in the superior pole of spleen. An magnetic resonance imaging was performed and re-

vealed 93x85 mm cystic lesion with septae in the superior pole of spleen and a heterogenic mass on the medial side of right rectus muscle (Fig. 1). Due to suspicion of hydatic cyst disease, serologic examination was performed and resulted as negative. In accordance with physical examination and radiological assessment, surgery was indicated due to huge splenic cyst and abdominal wall lesion. Laparoscopic splenectomy and mass excision from the anterior abdominal wall were performed. The patient was discharged without complications on the third postoperative day. Pathology was reported as endometriosis for the mass from the anterior abdominal wall. Macroscopic examination of the spleen revealed 110x100x98 mm cystic mass at the superior pole of spleen. Inner surface of the cyst was yellow-tan colored with smooth texture. Microscopic examination showed that cyst had no epithelial lining and there were two foci of endometriosis between the hyalinized wall layers near the splenic parenchyma (Fig. 2). Immunohistochemical study showed estrogen and progesterone receptor positivity (Fig. 2). Pathologic diagnosis was reported as "cystic mass with no epithelial lining, foci of endometriosis in the cyst wall". During the follow-up, there was no problem.

Informed consent was obtained from the patient and the Institutional Review Board approved the study (approval no: 2020-01/8).

DISCUSSION

Although extrapelvic endometriosis was reported in various organs before, spleen was mentioned as refractory for endometriosis. This case, as the fourth in the field since 2020, is valuable and unique by supporting the previous papers on splenic endometriosis and strengthening the existing literature [8–10]. The evaluation of the 3 cases with our case will guide us in assessing clinical presentation of splenic endometriosis and its interpretation. The demographic, clinical and pathological data of the four cases are given in Table 1.

Studies showed that endometriosis has multifactorial nature and there are several factors contributing to susceptibility of individuals such as genetic alterations, immune impairments and environmental factors [3]. Hypothesis of natural killer cell dysfunction in endometriosis is thought and mentioned to be in the pathophysiology of the disease [11]. While our patient has no immunological disorder, our report reveals that endometriosis of the spleen may occur in immunocompetent individuals.

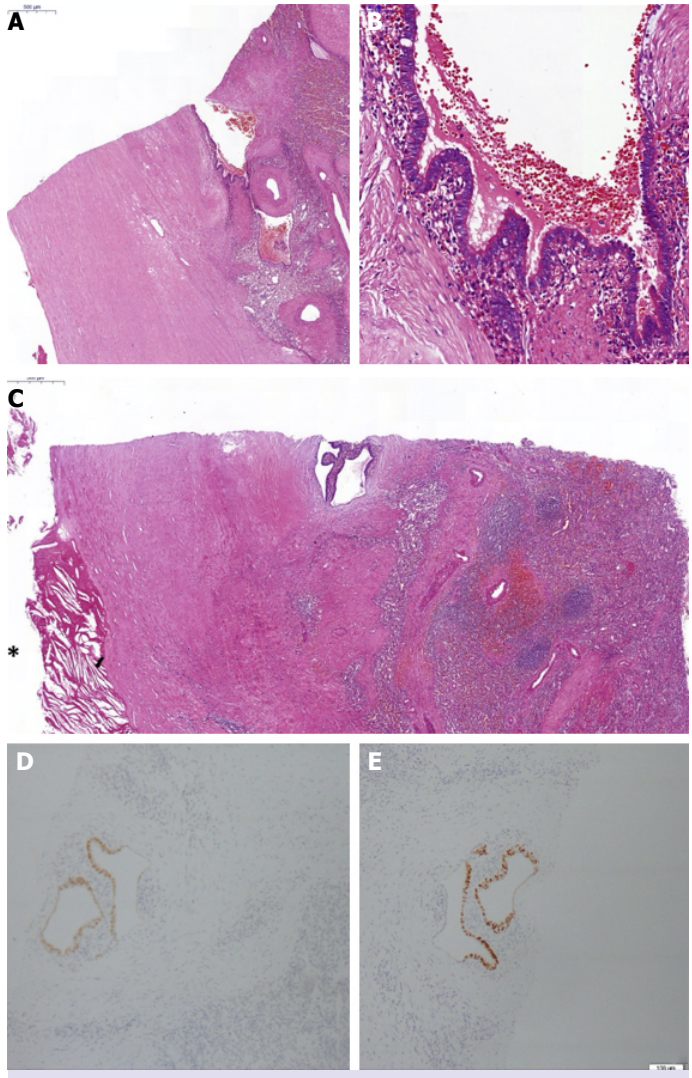

FIGURE 2. Endometrial glands in the cyst wall near the splenic parenchyma **(A)**; same glands with scant stroma in higher magnification **(B)**. Second focus of endometriosis near the splenic parenchyma, Asterisk denotes the lumen of the cyst **(C)** (H&E Stain). ER **(D)** (×100 µm) and PR **(E)** (×200 µm) positivity in the endometriosis focus.

Earlier in the literature, Samani et al. [12] reported that migration of endometriosis-derived cells to various organs, including spleen is possible in a murine model. Weyl et al. [10] reported the first case of splenic endometriosis in a patient with metastatic breast cancer. Coelomic metaplasia and vascular/lymphatic metastasis theories may be more coherent for extrapelvic implantations, including spleen as in this case.

As explained in another case report, splenic endometriosis was diagnosed in a patient with history of splenorrhaphy 10 years ago [8]. Authors postulated

Table 1. Demographic, clinical and pathological features of the cases

Cases	First case	Second case	Third case	Our case
Date of publication	September 2020	February 2022	December 2022	
Menopause	Pre-menopausal	Pre-menopausal	Pre-menopausal	Pre-menopausal
Patient age	54	36	26	31
Symptoms	Asymptomatic	Chronic left upper quadrant pain with postprandial nausea and vomiting	Bloating, and early satiety	Right lower quadrant pain
Preoperative imaging findings	13 cm compartmentalised fluid filled cystic structure with FDG uptake (SUV_{max}: 4.8)	8 cm multiloculated cystic mass with mural calcifications	10 cm splenic cyst - mass effect on stomach	9 cm cystic lesion with septae in the superior pole of spleen
Previous history of endometriosis	No	NM	No	No
Differential diagnosis	Breast cancer metastasis	Traumatic, iatrogenic, infectious, pseudocyst	traumatic, vascular, infectious	Iatrogenic, infectious
Surgery (laparoscopic/open)	Laparoscopic	Open	Laparoscopic	Laparoscopic
Length of stay at hospital	NM	NM	1	3
Pathology	Uncomplicated splenic cyst with chronic inflamatory changes, ER+, PR+, HER2-, PAX8+	Multiple cystic structures, glands lined by mucin secreting columnar epithelium	Chronic hemorrhage, PAX8 +, estrogen+, progesterone+	No epithelial lining + 2 foci of endometriosis between hyalinized wall layers near splenic parenchyma
Follow-up	2 years follow-up without problem	Lost to follow-up	NM	No problem in the follow - up

NM: Not mentioned.

the iatrogenic seeding mechanism as the pathophysiology for that case [4]. Since gynecological intervention history of the patient was non-remarkable, to us, it is not possible to agree with this interpretation. In our case, the mass on the anterior abdominal wall was likely caused by iatrogenic implantation due to previous cesarean section. There is also possibility for the same mechanism of seeding in splenic implantation but other theories such as coelomic metaplasia, vascular and lymphatic metastasis may also be coherent.

The last case in the literature reports incidentally found splenic endometriosis in a 26-year-old female patient who had type 1 DM and eosinophilic esophagitis [9]. After identification of a 10x9x9 cm splenic cyst in cross-sectional imaging, the patient had a laparoscopic splenectomy due to size and proximity to the splenic hilum of the cyst. The main complaints of the patient were bloating and early satiety and these were contributed to her eosinophilic esophagitis. Like our case, after resection of huge splenic cyst, final pathology revealed splenic tissue with an endometriotic cyst as characterized by endometrial glands and stroma unexpectedly. Also, the authors claimed that their patient had no history of endometriosis, had regular menstrual cycles and was nulliparous. In all three cases, also in our report, estrogen and progesterone receptor positivity were used as indicators of endometriosis.

As stated in current guidelines, surgical removal is the best option for the treatment of extrapelvic ab-

dominal endometriosis [13, 14]. On the other hand, if endometriosis is confirmed with biopsy and the patient has no symptoms due to endometriosis, clinical follow-up with routine ultrasound monitoring is sufficient [5, 14, 15]. Infertility and pain have been the major indications for operation in most cases. In our case, due to the patient's unidentified abdominal pain for two months and the complex cystic lesion in spleen, resection is done for all lesions possibly culprit. The patient's complaints disappeared immediately after surgery. Furthermore, intraparenchymal hemorrhage and subsequent spontaneous perforation of the spleen due to presence of endometrial tissue might be an emergent and devastating clinical presentation.

In conclusion, the idea of resistance of the spleen against endometriosis became outdated with this case report as the fourth case in the literature. Therefore, splenic endometriosis is getting out of being a rare occasion and can be thought of as a differential diagnosis for lesions of spleen in women with unidentified cyclic abdominal pain.

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Informed Consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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