

Pelvic Splenosis after a traumatic splenectomy: A case report and literature review

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Abstract

Splenosis is a rare benign condition that ectopic spleen tissue auto-transplant after trauma or splenectomy. Pelvic splenosis is often misdiagnosed as gynecological tumor, which results in unnecessary surgical treatment. In this article, a case of splenosis that was misdiagnosed as gynecological tumor prior to laparoscopic resection is presented, similar cases are also analyzed using literature review.

Keywords: splenosis, pelvic mass; splenectomy; laparoscopy

Introduction

Splenosis, an ectopic auto-transplantation of spleen tissue, is a benign and mostly asymptomatic condition that usually occurs after trauma or splenectomy. It is most commonly seen in abdominal cavity following pelvic cavity, rare cases such as thoracic, appendiceal, pleural splenosis were also reported. The lesions usually appear to be multiple round well-circumscribed purple-blue nodules, which can easily be misdiagnosed as tumor. Here, informed by patient's written consent, we present a case of pelvic splenosis after a traumatic splenectomy as well as review relative literature.

Case Presentations

A 24-year-old female underwent splenectomy for splenic ruptured due to car accident. On December 17 2019, she visited our hospital with the complain of disturbance of menstrual cycle for 2 months. Prior to the onset of her symptoms, her menstrual cycle was 7/28-32 days, then in the past two months, it was disordered and shortened to 6-7/15-20 days without presenting abdominal pain, distension and vaginal bleeding or other specific discomfort. No specific family history was acknowledged. No typical sign was detected in abdominal palpation and bimanual examination. Ultrasonography displayed a solid-cystic mass measuring 65 × 32 mm (Figure 1), multiple cavities could be seen in the cystic part of the mass, and the solid part was measured 34 × 23 mm, blood flow signals could be seen in the solid part but was absent in the cystic part under Color Doppler Flow Imaging (CDFI). An intrauterine mass, which was considered a endometrial polyp was also discovered. With the discover of the pelvic mass, we further examined serum tumor markers, CA199, CA125, AFP, CEA, HE4 level were within normal range.

Combining the clinical manifestations, we acquired, pelvic tumor was considered. After fully informed the patient about her condition and treatment plans, we performed laparoscopic exploration on December 18, 2019. During the surgery, we discovered scattered purple-blue masses with different sizes on the surface of the left ovary, the largest was about 1 × 1cm (Figure 2a), a 6 × 5 cm oblate purple-blue mass with wide and tortuous vessels was seen on the surface of the ureter on the right pelvic wall (Figure 2b). There was no other obvious abnormality observed in pelvic and abdominal cavity. Hence, we resected the abnormal pelvic masses. Intraoperative fast-frozen pathology suggested non-cancerous tissue. Postoperative paraffin embedded pathology reported spleen tissue (Figure 3). The patient recovered well after the operation without surgical complications, and was discharged from hospital 5 days after the surgery, she has no symptoms by our latest follow-up on 20 June 2020, however, her menstrual cycle was not restored to normal before oral contraceptive treatment.

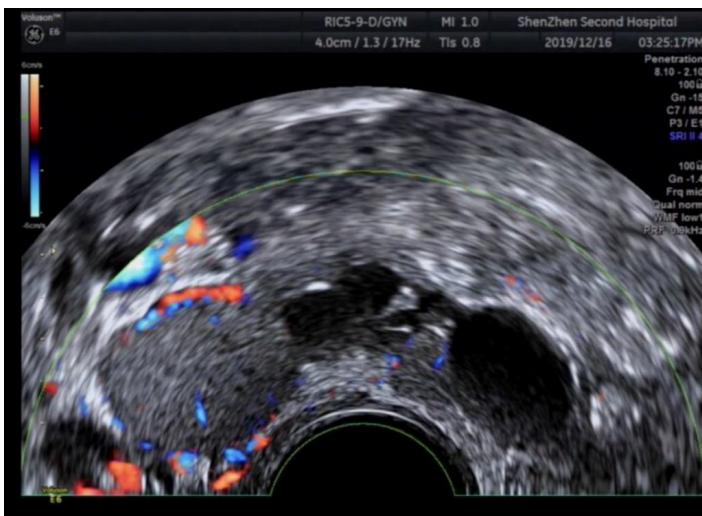


Figure 1. Ultrasonography of the right adnexa area. A mixed solid-cystic echoic mass region is measured 65 × 32 mm, the cystic part consists multiple cavities, the solid part is about 34 × 23 mm, CDFI: blood flow signal can be seen in the solid part but is absent in the cystic part.

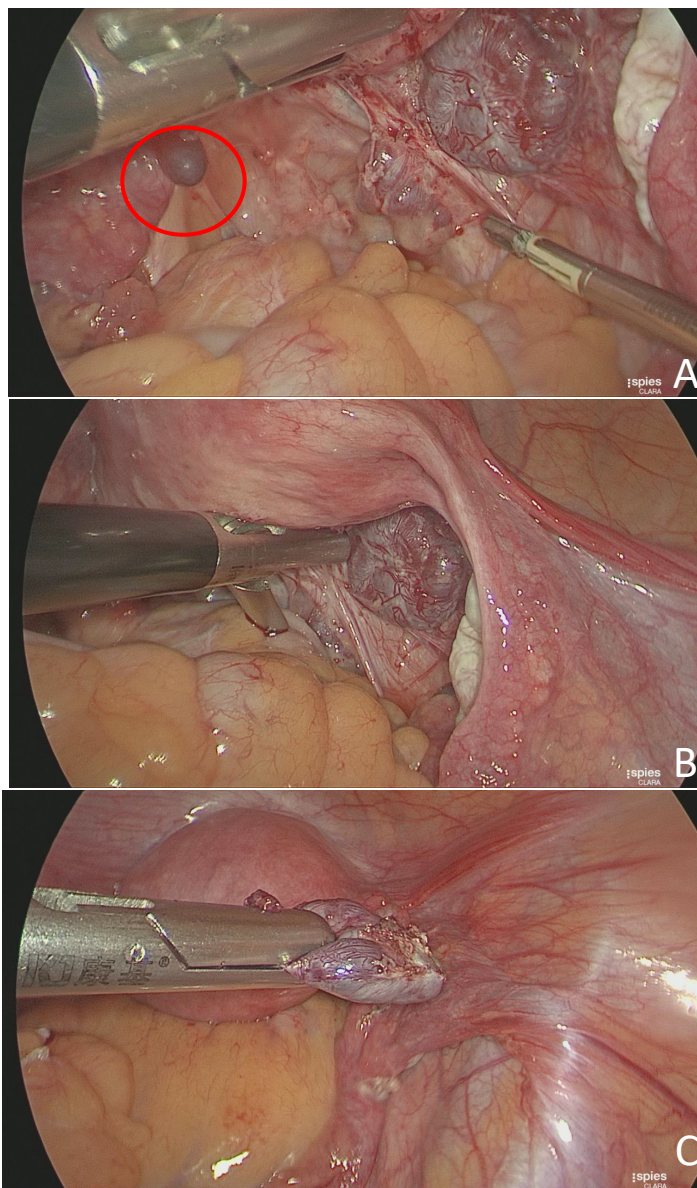


Figure 2. Laparoscopic exploration of the pelvic cavity. (A) Purple-blue mass (in red circle) on the surface of the left ovary. (B) Purple-blue mass on the surface of the right pelvic wall. (C) Longitudinal section of the mass after tumor removal.

Discussion

Ectopic splenic tissue exists in the body can be categorized into three forms: accessory spleen, splenogonadal fusion and splenosis [1]. Accessory spleen is a relatively common condition, it usually locates independently but supplied by splenic artery like the principle spleen. Splenogonadal fusion is a rare congenital malformation. Splenosis is also known as Ectopic Spleen Autotrasplantation (ESAT), usually occurs after splenic traumatic or splenectomy, and most commonly seen in abdominal cavity and pelvic cavity [2].

Spenosis is a benign, usually asymptomatic lesion. It is mostly detected via accidental physical or ultrasonic examination; however, different symptoms may appear based on the locations of the implants: abdominal pain caused by omentum infarction, obstruction, gastrointestinal bleeding caused by intestinal wall invasion, hydronephrosis caused by pressure on the ureter and etc

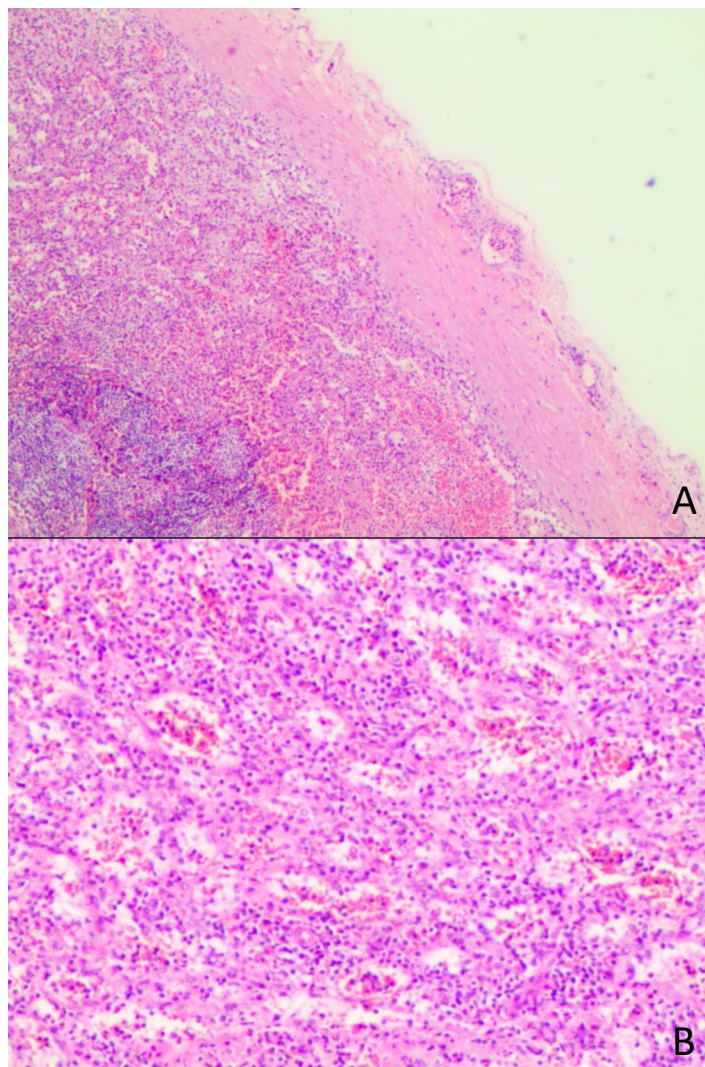


Figure 3. H&E staining. (A) Spleen tissue enclosed by a well-developed fibrous capsule (4X). (B) Spleen tissue (10X).

[2]. In the past 40 years, there were about 27 articles (in English), including a total of 29 cases, on pelvic and abdominal splenosis [2-28]. The average age of these cases is 42.6 years old, means of diagnosis include medical imaging, puncture biopsy, biopsy during laparoscopic exploration. Table 1 shows the characteristics of all these cases. Out of all cases, the most common symptom is pain (15/29), about half of it is serendipitous (14/29), implantation near the rectum caused gastrointestinal symptoms in 2 case [8,19]. Abnormal menstruation were detected in 5 cases [11-13,18,25], one of which had uterus myoma [13], and another had uterine adenomyoma [25], no abnormality was found in uterus and adnexa areas in the rest of the 3 cases. There is no clear evidence of correlation between splenosis and menstruation cycle. Infertility was reported in one case who underwent surgical treatment for fallopian tube blockage, in this case, pelvic inflammation caused by splenosis was considered a possible cause for her infertility [5].

Pelvic splenosis usually mimics pelvic tumors, making preoperative diagnosis difficult. This results in unnecessary surgical trauma, hence, differential diagnosis prior to the surgery is crucial. By far, the most accurate method is Tc-99 m heat-damaged erythrocytes and Tc-99m sulfur colloid scintigraphy or In-111 labelled platelets using PET-CT [25]. However, the unaffordable

Table 1. Pelvic splenosis; Literature review

Author	Year	Sex(age)	Symptom	Location(characteristic)	Complica-tion	Examination	Treatment
TODD H; OVERTON J.D.et al.	1982	female(34)	Abdominal pain	Pelvic (multiple)	none	biopsy	Laparotomy hysterectomy
KENNETH, SIRINEK et. al	1984	male(25)	Abdominal pain	Pelvic cavity (9*4*4.5cm)	Patrial bowel obstruction	bariumenema, abdominal CT	Laparotomy nodule resected
Linda M.Matonis et al.	1995	female(19)	none	abdomen and pelvis (multiple, 0.5-3cm)	none	Laparoscopy	laparoscopy nodule excis
Peer zitzer et al.	1998	female(35)	Abdominal pain	uterus (1cm) leftovarian(3*4cm)left cul-de-sac (7, 0.5-2cm) serosal surface of the rectosigmoid (2)	none	intravigenalultrasound, CA125	Left salpingo-oophorectomy
Murat Celiloglu et al.	2004	female(21)	menstrual irregularity, pelvic pain and mild dysmenorrhoea	Pelvic cavity(multiple, the most 6*4cm)	none	Abdominopelvic ultrasonography, CA125	laparotomy Nodules excise
B Vydianath et al	2005	female(61)	recurrent postmenopausal vaginal bleeding	Right ovarian(2cm)	none	Vaginal ultrasound	hysterectomy and bilateral salpingoophorectomy
Yasemin Tasci	2005	female(34)	abnormal vaginal bleeding, menometrorrhagia	colonic serosa, small , ileocecal region (multiple, 2-5cm)	none	Vaginal ultrasound, CA199, CA125, CEA, CA153, AFP, HCG,	hysterectomy, right salphengectomy, appendectomyand nodule excis
K. M. SARRAF et al.	2006	female(38)	bilateral lower abdominal pain	pelvis, omentum and the surfaces of the large and small bowel (multiple, 1-4cm)	none	transvaginal ultrasonnd, CA125	Laparotomy nodule excis
Jorge C. Ribeiro Et al.	2006	male(42)	none	Pelvic cavity (multiple, the most 3.7*2.8cm)	none	ultrasound, CT, MRI, DHL, α -FP, β -HCG	Laparotomy nodule excis
Richard D.Fremont et al.	2007	male(54)	left-sided pleurisy	left-sided pleuralbased lesions, (multiple, the most 2cm)	none	chest x-ray, chest CT, heat-damaged red blood cell scan	Medicated analgesic therapy
Vaknin Zvi, et al.	2009	female(39)	right lower abdominal pain	Right ovarian(5cm)	none	β -human chorionic gonadotrophin urine test, Transvaginal and abdominal ultrasonnd, contrast-enhance CT, laparoscopy biopsy	Conservative treatment
H. Talati and Jasim Radhi	2010	female(53)	acute right lower quadrant abdominal pain (hydrosalpinx on right fallopian)	Left ovarian serosa(5mm)	none	Urine examination, abdominal ultrasound	bilateral salpingoophorectomy

Enobong Ekong et al.	2011	female(35)	menometrorrhagia	abdomen and pelvis (multiple, the most 4*3cm)	none	ultrasound, abdominal CT	laparoscopic hysterectomy with removal of the mass
V. Gincu, et al.	2011	female(74)	intermittent constipation	rectal fold (2.5cm)	none	contrast-enhance CT, MRI, Colonoscopy	laparoscopy nodule excise
Sinan Akay et al.	2012	male(23)	none	retrovesical and pelvic regions (multiple)	none	Abdominal ultrasound, contrast-enhance CT, MRI and Tc-99m sulfur colloid scintigraphy	none
Antonio Mollo et al.	2011	female(28)	infertility (bilateral tubal occlusion)	Douglas fossa(multiple, the most 5cm)	none	ultrasound, CA125	Laparoscopy biopsy
R.Duivenvoorden et al.	2013	male(44)	none	left upper abdomen (2.5*3.5cm), pelvic cavity(7.6*4.3cm)	none	abdominal CT	none
Denisse Anrique et al.	2013	female(32)	chronic relapsing pelvic pain	douglas fossa, the bowel surface	none	Vaginal ultrasound, rectal endoscopy, MRI	laparoscopy nodule excise
Sue Yeon Park et al.	2014	female(24)	lower abdominal pain	pelvic cavity (45*30mm)	none	ultrasound, CA125, CA199	laparoscopy nodule excise
Deanna Gerber et al.	2015	female(56)	none	upper abdomen, posterior to the cervix, on the left uterine fundus and adjacent to the left ovary. (multiple, the most 4.5*2.8*4.4cm)	none	ultrasound, CT, MRI	abdominal hysterectomy, bilateral salpingo-oophorectomy, appendectomy, partial omentectomy and removal of rectal nodules
RAQUEL LAMEIRAS et al.	2017	female(54)	pelvic pain and vaginal bleeding	Pelvic cavity(5.2cm)	none	CA125, CA199, HCG, AFP, Endovaginal ultrasound, MRI	hysterectomy with bilateral adnexectomy
Hélder Ferreira et al.	2017	female(40)	severe dyspareunia and dyschezia	douglas fossa	none	ultrasound, MRI	laparoscopy nodule excise
Yasmeen K. Tandon et al.	2018	male(64)	none	between the bladder and rectum(5.2*2.9cm)	none	CT, MRI, The sulfur colloid scan	none
		male(53)	none	left seminal vesicle (3.5*2.1cm)	none	MRI	radical robotic prostatectomy (prostate cancer)
		male(65)	none	left subphrenic region, peritoneal reflection adjacent to the seminal vesicles, perihepatic region, and in the right inguinal canal (multiple)	none	CT	none
Zhong Jia et al.	2019	female(49)	none	douglas fossa (multiple, 1cm-4cm)	none	Vaginal ultrasound, contrast-enhanced CT	none
Ross Kruger et al.	2019	male(56)	none	rectosigmoid junction (4.3*2.7cm)	none	MRI, a contrast-enhanced CT scan of the abdomen and pelvis, contrast-enhanced ultrasound	none

Georgia Karpathiou et al.	2019	female(46)	intense pain of the right iliac fossa	Right ovary(2cm)	none	Pelvic CT	laparoscopy nodule excise
Darshan Gandhi et al.	2020	female(36)	intermittent left flank pain (left-sided gross hydronephrosis)	right hepatic lobe (5cm)	none	ultrasound, AFP, MRI	computed tomography (CT)-guided core biopsy

cost, long awaiting time and radiation making it unpractical to popularize. A recently reported case used tissue-specific contrast agent Superparamagnetic Iron Oxide (SPIO) under MRI to successfully detect splenosis, but its sensitivity and specificity is unfavorable [25]. The European Federation of Societies for Ultrasound in Medicine and Biology (EFSUMB) recommend use Contrast Enhanced Ultrasound (CEUS) rather than PET-CT to diagnosis splenosis [27]. Splenic rupture caused by trauma has become one of the most common cause for emergency surgery due to car accident. Given that pelvic splenosis is often misdiagnosed, and as above mentioned, practical approaches to determine the disease is lacking, it might be more important to pay attention to the medical history when encounter patients with pelvic tumor.

With regard to treatments, whether the ectopic splenic tissue should be excised remains controversial. Some scholars believe that ectopic splenic tissue has certain immune activity after splenectomy, which can prevent severe infection after splenectomy [3]. However, after reviewing and analyzing the difference between the structure of the heterotopic and normal spleen, some suggested that the heterotopic spleen may not be able to serve the same immunologic function. Fatal pneumococcal and meningococcal septicemia infection occurred in many cases despite the existence heterotopic spleen [6]. Although the purpose of reserving the ectopic spleen is debatable, no evidence of harm is observed among patients with asymptomatic splenosis. Hence, secondary surgical injury should be avoided among these patients.

In summary, pelvic splenosis is rare, and preoperative diagnosis is difficult, often misdiagnosed as pelvic mass. Therefore, gynecologists should fully consider the possibility of spleen implantation in combination with the medical history of patients with pelvic masses and prior history of spleen rupture or splenectomy. For asymptomatic patients, medical observation is preferable, and for patients with symptoms caused by splenosis, surgical resection can be performed.

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Conflict of Interest

The authors declare that there are no conflict of interests.

Ethical Approval

This article does not contain any studies with human participants or animals performed by any of the authors

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