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Case report

Cecal splenosis mimicking gastrointestinal stroma tumor. Case report and review of the literature

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ABSTRACT

Introduction: Abdominal splenosis is a rare condition where autotransplanted ectopic spleen tissue is found in the abdominal cavity after spleen injury or splenectomy. While abdominal splenosis is mostly described as asymptomatic, bowel splenosis can present with abdominal pain, obstipation or gastrointestinal (GI) bleeding. Scarce information on bowel splenosis exists and high index of suspicion is needed for diagnosis. We present the case of a patient with abdominal pain and a cecal mass mimicking gastrointestinal stroma tumor (GIST) found to have bowel splenosis after laparoscopic resection.

Presentation of case: A 45-year-old female was evaluated for a 6-month history of abdominal pain in right-lower quadrant. She had past medical history of blunt abdominal trauma and splenectomy 35 years before symptoms. An abdominal contrast-enhanced computed tomography (CT) reported a 4.2 × 4.6 × 4.6 cm solid mass located in the antimesenteric border of the cecum, with calcifications and arterial enhancement. Colonoscopy found no epithelial lesions. A diagnostic laparoscopy was done, and resection of the mass achieved. Transoperative histopathological diagnosis revealed ectopic spleen tissue.

Discussion: It is difficult to know whether abdominal pain in bowel splenosis is actually triggered by splenosis itself or by an unrelated cause, making diagnosis incidental. There are no particular imaging findings suggestive of splenosis, and surgical resection is often done under uncertain diagnosis.

Conclusion: Bowel splenosis could present as abdominal pain variable time after spleen injury or splenectomy. Clinical features and imaging characteristics are nonspecific and similar to those of GIST. Most cases of splenosis are diagnosed after surgery.

1. Introduction

Abdominal splenosis is a benign condition where autotransplanted ectopic spleen tissue is found in the abdominal cavity after traumatic spleen injury or splenectomy [1]. Although the exact incidence is unknown given a frequent asymptomatic course, authors claim it to be above 70% in patients with traumatic injury and splenectomy [2]. Seeding of splenic parenchyma in the abdominal cavity and hematogeneous spread of splenic pulp have been suggested as possible mechanisms of autotrasplantation [3]. Implants have been found on intra and extraperitoneal structures, such as small and large bowel, omentum, peritoneum, mesentery, diaphragm, stomach, liver, pericardium, among others [1]. While this entity could arise as an incidental finding on image studies or surgical procedures for unrelated causes, symptoms such as gastrointestinal (GI) bleeding, constipation, and abdominal pain have been reported regarding lesions on the gastrointestinal tract [4,5]. A high index of suspicion is needed for diagnosis and it is not uncommon to misdiagnose implants on the small and large bowel as gastrointestinal neoplasms such as gastrointestinal stroma tumor or lymphadenopathy [6,7].

We present the case of a 45-year-old female who presented to the emergency department with abdominal pain. She had past medical history of blunt abdominal trauma and splenectomy in childhood. Imaging results reported a cecal mass and gastrointestinal neoplasia was suspected. Laparoscopy was performed and splenosis was confirmed by transoperative analysis. Patient was managed in a private care hospital. This case report is compliant with the SCARE and PROCESS guidelines [8,9].

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2. Case report

A 45-year-old-female presented with a 6-month history of abdominal right-lower-quadrant pain. During the last two weeks pain had intensified reason for which she sought medical attention. Patient denied pain irradiation, nausea, vomiting, weight loss, change in bowel habits, or transrectal bleeding. She had past medical history of splenectomy due to blunt abdominal trauma 35 years before onset of symptoms. Physical examination revealed vital signs within normal limits. She had right-lower-quadrant tenderness, but no abdominal mass was palpable. Laboratory analysis was all normal (Table 1). A contrast-enhanced computed tomography (CT) revealed a 4.2 × 4.6 × 4.6 cm solid mass located in the antimesenteric border of the cecum, with central calcifications and arterial sustained enhancement (Fig. 1). Colonoscopy was done and no intraluminal lesions where found. During cecal visualization there was difficulty at insufflation, suggesting an extrinsic or mural mass effect at this location. No biopsies where taken. The patient was programmed for a diagnostic laparoscopy.

During laparoscopy, a 5 × 5 cm mass was found adherent to the serosal surface of the cecum and omentum (Fig. 2). Dissection was cautiously done with thermocoagulation energy. The specimen was extracted and sent for histopathological transopertive analysis (Fig. 3). Results confirmed normal splenic tissue without malignancy (Fig. 4).

The patient was discharged home the next day, without complications.

3. Discussion

Ectopic splenic tissue can be found as an accessory spleen, product of a congenital anomaly, or as an acquired condition named splenosis [3]. As opposed to an accessory spleen, splenosis usually has blood supply derived from surrounding tissue and could be found on any peritoneal or extraperitoneal structure [3]. Splenosis has been found in small and large bowel, omentum, peritoneum, mesentery, diaphragm, stomach, liver, pericardium, among others [1]. Fifteen cases of bowel splenosis have been published most of which have been misdiagnosed for gastrointestinal neoplasm (Table 2).

Age ranges from 9 to 74 years old, and most patents are male. This could reflect the incremented frequency of male patients suffering from abdominal trauma during teenage years. Splenectomy for trauma has been the most frequent risk factor, and sustains the physiopathological mechanism of splenosis, where splenic pulp is spilled from splenic tissue and seeds on intraperitoneal structures. Moreover, splenectomy for other causes, such as hereditary spherocytosis has been reported supporting this thesis [10].

Although splenosis has been mostly described as an otherwise asymptomatic entity, all 15 cases reported symptoms such as abdominal pain, obstipation, or gastrointestinal bleeding. Abdominal pain has been reported as the most frequent symptom in bowel splenosis, nevertheless, it is difficult to know whether pain was actually triggered by the ectopic splenic tissue or by an unrelated cause. Location of abdominal pain as to location of splenosis along the gastrointestinal tract helps clarify this issue. In our case, abdominal pain was located on the lower left quadrant, site where the mass was found. Pain could arise by external compression, infarction, bleeding or congestion of splenic tissue [1]. Gastrointesitnal bleeding occurs when splenic tissue is located within the bowel wall and through intestinal mucosae. Bowel obstruction caused by bridges and adhesions between splenic implants in mesentery and bowel wall has been reported [11,12]. However, bowel obstruction due to adhesions could be triggered not by the splenosis itself, but by a previous splenectomy, making splenosis an incidental finding after all [13].

Time from surgical past medical history to symptoms varies from 2 to 35 years. It would seem logic to think ectopic tissue size and/or location would influence time to symptoms, but no relationship has yet been described in the literature. Large bowel splenosis was more frequent than small bowel splenosis, and the largest ectopic tissue segment was found alongside the rectal wall in a patient with gastrointestinal obstruction [13]. As in this case, our patient presented symptoms 35 years after surgery.

CT scan in our case revealed a heterogenic cecal mass with arterial sustained enhancement. Imaging features of ectopic spleen tissue found on small or large bowel are nonspecific, most often presenting arterial enhancement, with isodensity in venous and portal phases. Many entities could be confused with splenosis such as metastasis disease, abdominal lymphoma, GIST, among others [1]. On MRI, ectopic splenic tissue is hypointense in T1 and hyperintense in T2 weighted images with heterogeneous contrast enhancement. Conventional MRI is usually not useful for differential diagnosis and was not a frequent technique on the approach of bowel splenosis [1].

Differential diagnosis is broad, and misdiagnosis for gastrointestinal neoplasms (particularly GIST), and lymphadenopathy have been reported [4,6,7]. As reported by Chetta et al. GIST can be found throughout the GI tract, 60% on stomach, 30% on small bowel, 4% on colorrectum [14–16]. Most are asymptomatic, and thus discovered incidentally; symptoms arise as they increase in size and are nonspecific, ranging from abdominal pain, nausea, weight loss to obstruction and GI bleeding [16]. Imaging features are highly variable. GIST arising in small bowel are commonly exophytic, may contain calcifications and present arterial enhancement [16]. Our patient presented abdominal pain and image studies reported an exophytic mass with calcifications and arterial enhancement, properties found in GIST. Conventional imaging techniques are not able to differentiate splenosis from other gastrointestinal tumors easily, however scintigraphy with technetium-99 m heat-denatured red blood cells has been reported as a reliable technique to diagnose splenosis [10,17]. As in this review, most studies reported diagnosis after surgical procedures. In patients in need for surgery because of GI obstruction or bleeding, a prior scintigraphy could be useful for surgical planning. In this case a transoperative histopathological diagnosis was made, and further treatment was not needed.

Table 1
Laboratory analysis at admission.

	Value	UM	Normal range
Hemoglobin	14.4	g/dL	13.2–18
Hematocrit	42.3	%	38.4–52.4
White blood cells	11.5	x10^3/ul	4.5–11
Platelets	326	x10^3/ul	150–420
Glucose	90	mg/dL	60–100
Creatinine	0.62	mg/dL	0.7–1.3
Blood urea nitrogen	14	mg/dL	8.9–25.7
Albumin	4.4	g/dL	3.5–5.0
AST	14	U/L	5–34
ALT	10	U/L	0–55
LDH	116.9	U/L	140–271
ALP	88.1	U/L	125–220
Amilase	52.5	U/L	25–125
INR	0.95		0.9–1.3

AST: aspartate aminotransferase, ALT: alanine aminotransferase, LDH: lactate dehydrogenase, ALP: alkaline phosphatase, INR: international normalize ratio.

4. Conclusion

We present the case of a patient with large bowel splenosis who had abdominal pain 35 years after splenectomy for trauma. Imaging techniques reported nonspecific characteristics and colonoscopy was negative for epithelial lesions. A GIST was suspected. Laparoscopy and mass resection were done and splenosis was diagnosed transoperatively. No further treatment was needed.

Patients with bowel splenosis can present abdominal pain, being this the most frequent symptom. Time from splenectomy to symptom presentation varies from 2 to 35 years and no relation to mass size has been described. Imaging techniques are nonspecific and could be

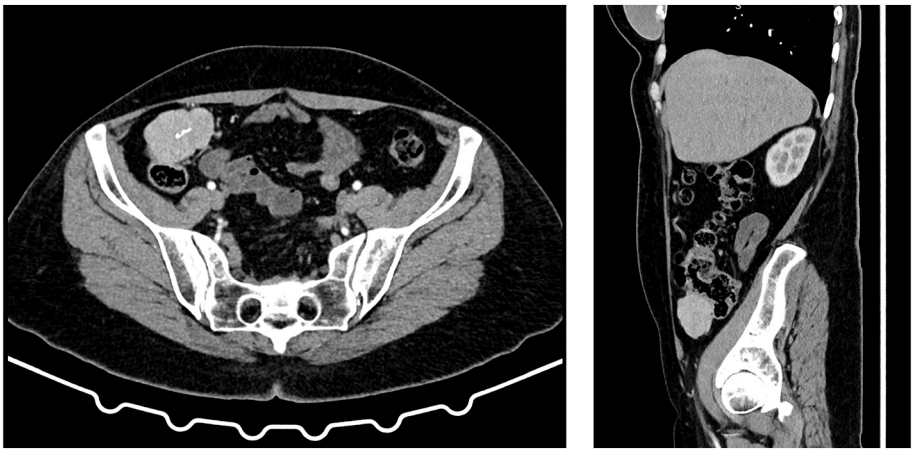

Fig. 1. 4.2 × 4.6 × 4.6 cm solid lesion located in the antimesenteric border of the cecum, with central calcifications and arterial sustained enhancement.

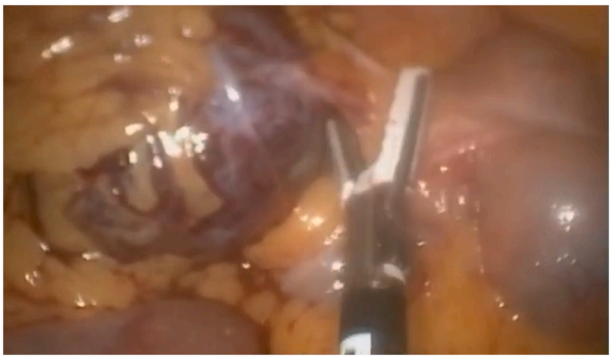

Fig. 2. 5 × 5 cm mass was found adherent to the serosal surface of the cecum and omentum. Disection was done for removal.

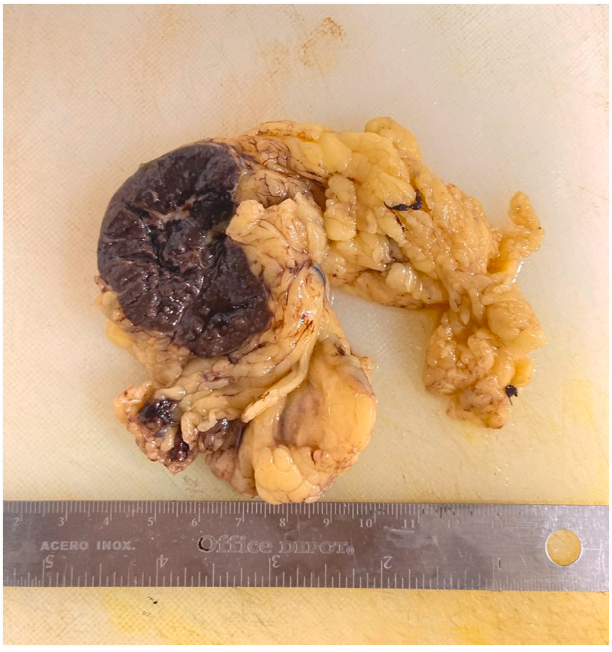

Fig. 3. Brown, lobulated 5 × 4 cm mass with congestive surface and hemorrhagic areas, surrounded by adipose tissue. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

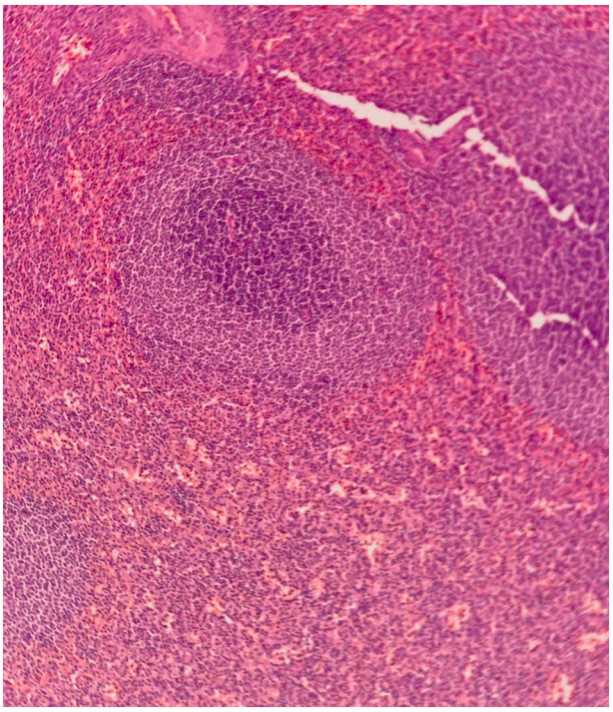

Fig. 4. Red and white pulp typical of splenic tissue with splenic sinusoids and lymphoid follicles. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

misdiagnosed for GIST. Most cases of splenosis are diagnosed after surgical resection.

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Ethical approval

N/a.

Table 2
Bowel splenosis review of the literature.

Author (ref)	Year	Age	Sex	PMH	Time[a] (years)	Symptoms	Location	Size (cm)	CT	MRI	Dx	Tx
Ksiądzyna [1]	2011	54	Female	Splenectomy for trauma	42	Abdominal pain	Ileal omentum	1.7 cm	–	–	–	Surgery
Obokhare et al. [4]	2012	41	Male	Splenectomy	2	Abdominal pain, obstipation, GI bleeding	Descending colon	6 cm	–	–	Lymphadenopathy	Surgery
Younan et al. [5]	2015	36	Male	Splenectomy for trauma	5	Abdominal pain, obstipation	Small bowel	3 cm	Arterial enhancement	–	–	Surgery
Chorbińska et al. [6]	2020	57	Male	Splenectomy for trauma	30	Abdominal pain	Omentum adjacent to sigmoid	3 cm	Isodense	–	Lymphadenopathy	Surgery
Xiao et al. [7]	2017	40	Male	Splenectomy for trauma	10	GI bleeding	Splenic flexure colon	5 cm	Arterial enhancement	–	GIST	Surgery
Kravarusik et al. [10]	2007	12	Male	Splenectomy for hereditary spherocytosis		Anemia	Splenic flexure colon	4 cm	–	–	Splenosis	Tc-99
El-Helou et al. [11]	2020	43	Male	Splenectomy for trauma	30	Abdominal pain, vomiting, bowel obstruction	Small bowel		Arterial enhancement	–	–	Surgery
El-Kheir et al. [12]	2019	46	Male	Splenectomy for trauma	12	Abdominal pain, obstipation	Jejunum Ileum Mesosigmoid		Arterial enhancement	–	Splenosis	Surgery
Garaci et al. [13]	2009	23	Male	Splenectomy for trauma	5	Abdominal pain, obstipation	Anterior rectal wall	10 cm	Arterial enhancement isodense in venous and portal phase	–	Splenosis	Surgery
Famà et al. [17]	2016	68	Male	Splenectomy for trauma	25	GI bleeding	Jejunum	1.6 cm	Arterial enhancement	–	Splenosis	Tc-99
Arena et al. [18]	2018	58	Male	Splenectomy for trauma	30	GI bleeding, bowel obstruction	Ileum	3 cm	Arterial enhancement	–	Small bowel neoplasia	Surgery
Gincu et al. [19]	2011	74	Female	Splenectomy for trauma	18	Obstipation	Rectosigmoid colon	2.5 cm	Arterial enhancement	Hypointense T2	Colorectal neoplasm	Surgery
Sato et al. [20]	2007	9	Female	Splenectomy for hereditary spherocytosis	4	Abdominal pain, vomiting	Ileal Mesenterium Sigmoid		–	–	–	Surgery
Abeles et al. [21]	2003	54	Male	Splenectomy for trauma	30	Abdominal pain, vomiting, GI bleeding	Jejunum	3 cm	Arterial enhancement	–	Lipoma	Surgery
Schenkein [22]	1995	65	Male	Splenectomy for trauma. Hodgkin disease	33	Pleuritic pain	Anterior mediastinum Sigmoid colon Descending colon	6 cm 4 cm 2.5 cm	Heterogeneous lobulated mass Hypodense and arterial enhancement	–	Hodgkin disease	Surgery

PMH: past medical history, CT: computed tomography, MRI: magnetic resonance imaging, Dx: diagnosis, Tx: final diagnosis, Tc-99: tecnesium-99.
[a] Time from splenectomy to symptoms.

Informed consent

An informed consent was sign by the patient.

Author contribution

Study concept, data collection, data analysis, writing the paper.

Guarantor

Alberto Riojas Garza.

Declaration of competing interest

None.

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