American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2020; 21: e921685
DOI: 10.12659/AJCR.921685

Received: 2019.11.25
Accepted: 2020.01.02
Available online: 2020.01.24
Published: 2020.04.17

Splenosis of the Mesoappendix with Acute Appendicitis: A Case Report

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Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 40-year-old
Final Diagnosis: Abdominal splenosis and acute appendicitis
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare co-existance of disease or pathology

Background: Splenosis is a benign condition involving the auto-transplantation of splenic tissue at various locations, resulting from splenic injury or splenectomy.

Case Report: A 40-year-old male, with a history of remote exploratory laparotomy with splenectomy secondary to blunt abdominal trauma, presented with symptoms consistent with acute appendicitis, which was subsequently confirmed by computed tomography scan of the abdomen that further demonstrated the presence of multiple abdominal nodules, one of which was adjacent to the appendix. A laparoscopic appendectomy was then performed along with resection of the nodule located in the mesoappendix, which was confirmed to be a splenic tissue based on histopathological examination.

Conclusions: Abdominal splenosis is not an uncommon condition in patients with a history of splenic injury. However, the involvement of the mesoappendix, which may or may not contribute to acute inflammation of the appendix, is very rare.

MeSH Keywords: Appendicitis • Splenectomy • Splenosis

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/921685

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Background

Splenosis is a benign condition that is defined as an auto-transplantation of viable splenic tissue onto a vascularized surface. This condition develops following splenic injury through traumatic rupture or splenectomy, which results in seeding or hematogenous spread of the detached splenic pulp cells. The auto-transplanted splenic cells derive their blood supply from adjacent structures and subsequently differentiate into mature splenic tissues. Although it is an uncommon condition, it affects approximately two-thirds of all patients with traumatic splenic rupture [1]. Herein, we present a case of a middle-aged male patient who presented with symptoms consistent with acute appendicitis; a subsequent computed tomography (CT) scan confirmed its diagnosis and further demonstrated multiple abdominal nodules, one of which was adjacent to the appendix. Subsequently, the patient underwent a laparoscopic appendectomy along with resection of the nodule in the mesoappendix. A histopathological examination of the resected tissue indicated the presence of encapsulated splenic tissue along with acute appendicitis.

Case Report

A 40-year-old male presented to the emergency department with a 2-day history of generalized abdominal pain that had localized to the right lower abdomen at the time of admission. He described the pain as sharp, constant, and being exacerbated by movement. Furthermore, he reported no bowel or urinary symptoms or subjective history of fever. The patient underwent an open splenectomy 20 years ago for a splenic injury resulting from a fall from height. He also gave a history of gastritis associated with *Helicobacter pylori* that was resolved successfully with triple therapy.

Upon presentation, he was afebrile, and his blood pressure, pulse rate, and respiratory rate were observed to be

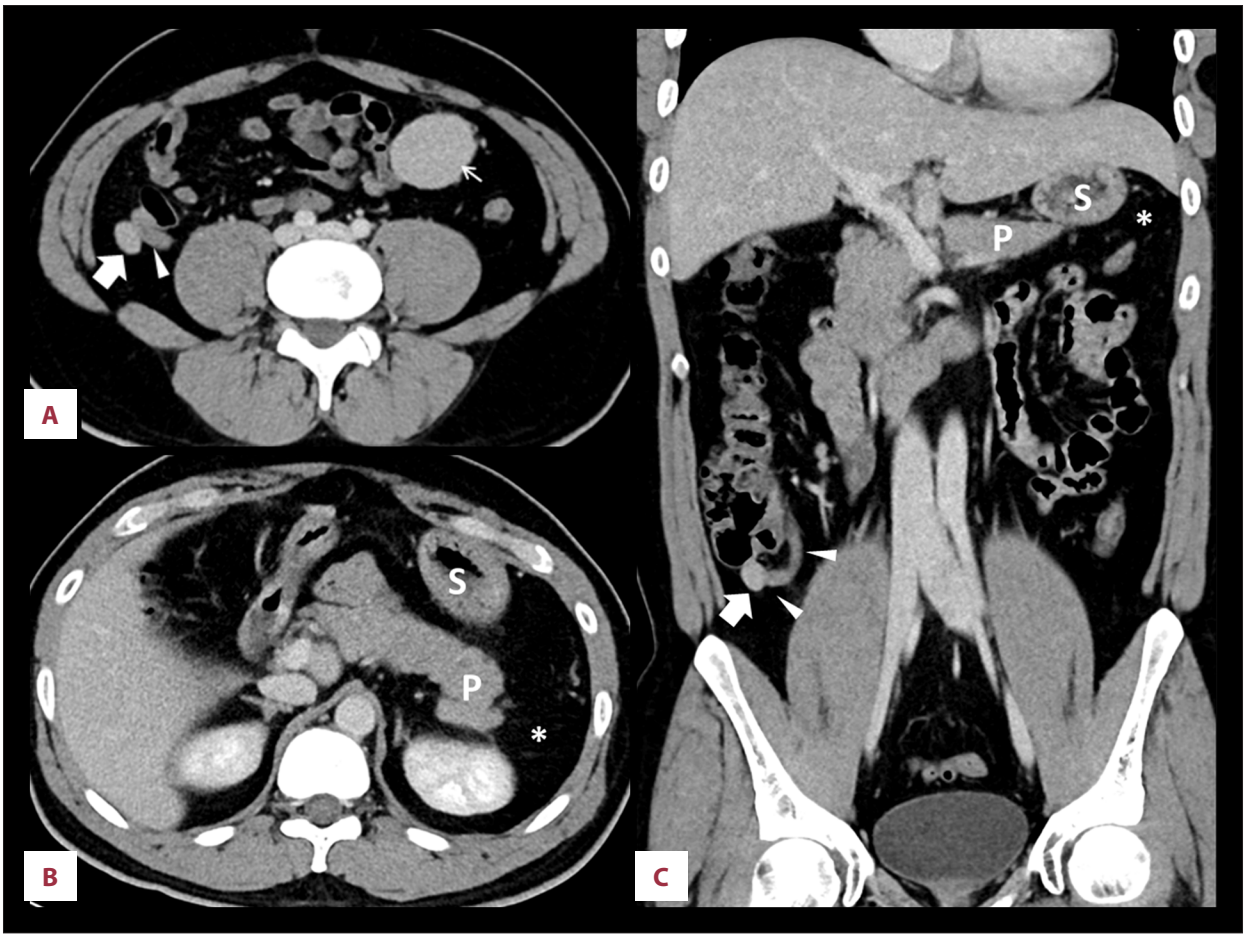

Figure 1. Contrast-enhanced abdominal computed tomography: Axial (**A:** at the lower abdomen; **B:** at the level of the splenic fossa), and coronal images (**C**). Note the enhancing soft tissue nodule (thick white arrow in **A** and **C**), representing the splenosis, adjacent to the appendix (arrow heads in **A** and **C**). The splenic fossa (asterisk in **B** and **C**) is empty. A large splenic nodule is also seen at the left lower quadrant (white arrow in **A**). Stomach (S), Pancreatic tail (P).

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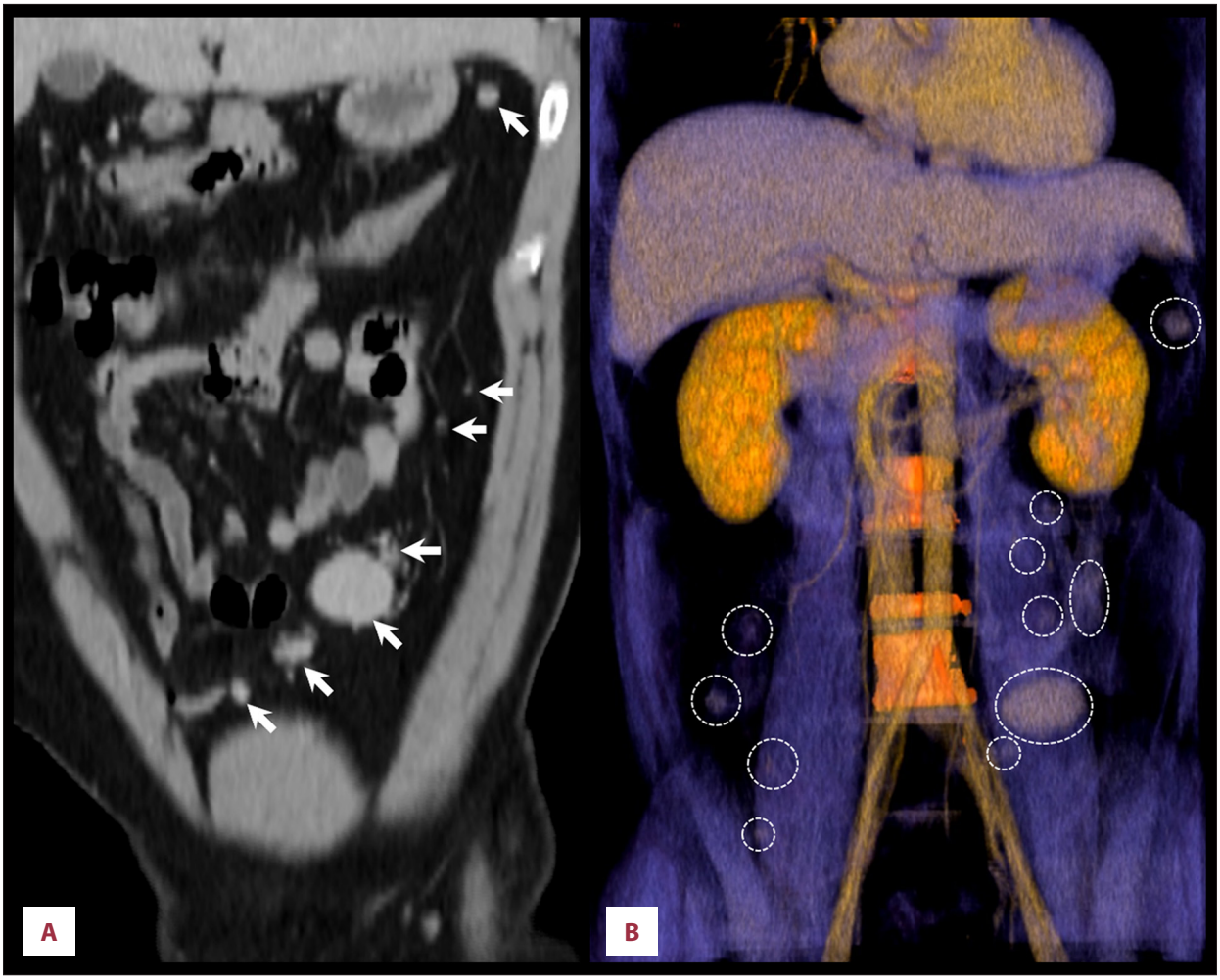

Figure 2. Multiplanar reconstruction (MPR) on coronal plane (**A**) and volume rendering technique (VRT) reconstruction demonstrate absence of native spleen and multifocal lesions (arrows in **A**, encircled in **B**) of different sizes and shapes scattered in the abdominopelvic region.

126/78 mmHg, 89 beats per minute, and 12 breaths per minute, respectively. The abdominal examination revealed a left paramedian scar from previous exploratory laparotomy. There was a localized tenderness in the right lower quadrant with no peritoneal signs. Additionally, the patient's blood analysis revealed a hemoglobin level of 15.9 g/dL, a leukocyte count of 14.4×10^3/mL with a left shift (88% neutrophils), and a platelet count of 497×10^3/mL, with his levels of urea, electrolytes, and liver enzymes found to be within the normal ranges. The urinalysis revealed no abnormalities. A peripheral blood smear showed no Heinz or Howell-Jolly bodies.

A CT scan of the abdomen was further performed to confirm the diagnosis of suspected acute appendicitis (Figures 1, 2). A mildly distended appendix with homogenous hyperenhancement of the appendiceal wall with no stranding of adjacent fat or extraluminal fluid collection was observed, which indicated early acute appendicitis. Additionally, a surgically absent spleen and the presence of multiple intra-peritoneal, well-defined nodules of soft tissue density suggested splenosis; the largest nodule was measuring 4.2×3.3×3.2 cm. Notably, one of the nodules was observed to be adjacent to the tip of the appendix.

Given the aforementioned clinical and radiological findings, the patient was then prepared for a laparoscopic appendectomy. The surgery was performed under general anesthesia with the patient placed in a supine position, and 3 ports were inserted to carry out the procedure. After establishing the pneumoperitoneum and introducing trocars, careful adhesiolysis was performed. The diagnostic exploration revealed multiple lobulated, and hypervascular nodules of variable size, which were consistent with splenosis (Figure 3). The largest nodule was embedded in the adhesion area of the previous laparotomy site. A resection of the edematous appendix along with the adjacent splenic nodule was successfully performed, and a histopathological examination showed normal splenic

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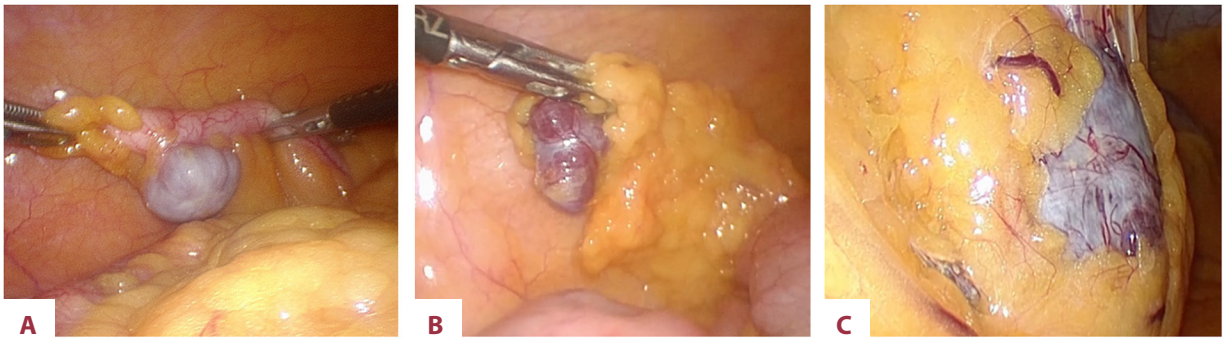

Figure 3. Laparoscopic view showing the multiple splenic nodules of variable sizes and shapes; notably, one of these nodules was located in the mesoappendix (**A–C**).

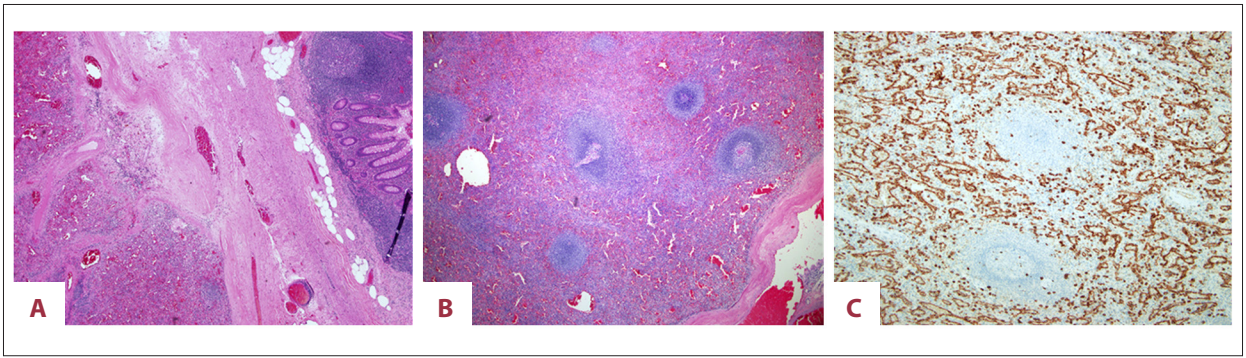

Figure 4. (**A**) Low power view (hematoxylin and eosin (H&E) stain, 4×) showing encapsulated ectopic splenic tissue (left) attached to the appendiceal wall (upper right). (**B**) Low power view (H&E stain, 4×) showing normal splenic tissue with white and red pulp. (**C**) Strong expression of CD8 by endothelial cells lining the sinusoids is shown, a feature that is unique to splenic-type endothelium [31] (CD8 immunohistochemical staining, 100×).

tissue with red and white pulp (Figure 4) along with acute appendicitis. The patient tolerated the procedure well with no complications and was discharged on the second postoperative day with no active complaints during the follow-up visit.

Discussion

Splenosis was first described by Albrecht in 1896 and the term itself was coined by Buchbinder and Lipkopff in 1939 [2]. A distinction must first be made here between accessory spleen, which is another type of ectopic splenic tissue, and splenosis (Table 1) [3].

While the typical site for splenosis is the abdomen, it is known to also occur at various locations. Abdominal splenosis, for instance, has been observed on the serosal surfaces of the small intestine and colon, mesentery, greater omentum, parietal peritoneum, undersurface of the diaphragm, stomach, pancreatic tail, and even inside the liver and the kidney. A study by Lin et al. demonstrated that the left upper abdominal quadrant is the most frequent site of splenosis [4]. Additionally, diaphragmatic rupture appears essential for the development of thoracic splenosis that allows for the seeding of splenic cells into the thoracic cavity resulting in solitary or multiple pleural-based nodules. However, a few cases of thoracic splenosis without diaphragmatic rupture have also been reported [5]. It is estimated that thoracic splenosis has been observed in one-fourth of abdominal splenosis cases. Splenosis can also develop in the pelvis and the pelvic organs. The hypothesis in which splenic pulp cells may survive after seeding has also been demonstrated in experimental studies; however, it is insufficient to explain the occurrence of splenosis in certain exceptional locations, including the brain [6].

As the vast majority of cases are asymptomatic and are incidentally identified through imaging, the time between the incidence and development of splenosis is still unclear. However, the average interval between splenectomy and development of splenosis has been reported to be approximately 2 decades [7]. The site and size of the lesions also influence the clinical presentation of splenosis. The nodules are typically small in size due to their limited blood supply. They are generally less than 3 cm in diameter [8]. However, nodules may grow as large as

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Table 1. Comparison between splenosis and accessory spleen.

		Splenosis	Accessory spleen
Etiopathogenesis		**Acquired:** direct seeding or hematogenous spread of splenic pulp cells following splenic injury	**Congenital:** failure of fusion of mesenchymal cells during embryonic development
Number		Usually multiple	Usually solitary
Size		Usually small	Varies
Location		Varies	Near spleen
Histology	Capsule	No smooth muscle component	Elastic muscular capsule
	Hilum	Absent	Present
	Trabecular Structure	Less prominent	More prominent
Blood supply		Parasitized	Splenic artery

13 cm in diameter [9]. In our case, the size of the nodules was consistent with splenosis. Splenosis might be misdiagnosed as benign or malignant neoplasms such as metastatic disease, and this may lead to unnecessary invasive procedures.

Abdominal splenosis may also present clinically with abdominal pain, intestinal obstruction, and hydronephrosis and unusually with gastrointestinal bleeding. Acute abdominal pain may develop from splenosis torsion and infarction. Intussusception has also been described as an etiology of abdominal pain in a patient with abdominal splenosis [10,11]. Hydronephrosis may develop as a consequence of external compression of ureters by the splenic nodules [12]. Adhesive bands of splenic nodules could result in intestinal obstruction or acute appendicitis. Splenic implant hematoma following trauma may also occur [13]. Gastrointestinal bleeding is specifically attributed to the presence of poorly formed capsule of splenosis which predisposes it to a spontaneous or traumatic rupture, resulting in occult or massive bleeding [14,15]. Similarly, thoracic splenosis may present clinically with chest pain, cough, or hemoptysis [16,17]. Pelvic splenosis may present in a clinical picture that mimics gynecological conditions, including endometriosis [18]. In our case, the clinical presentation of acute appendicitis may be related to the splenosis nodule despite that it did not cause any obstruction, as evident by imaging. We presume that the presence of splenic nodule in the mesoappendix, because of its parasitized blood supply, could have resulted in ischemia to the appendix. Such ischemia can cause acute appendicitis [19]. However, this is just speculation, as the coexistence could be merely incidental.

The mesoappendix is considered a rare site of pathology [9]. Very few cases of acute appendicitis have been reported in association with abdominal splenosis [20–23]; however, only 2 cases of splenosis involving the mesoappendix has been described [23,24].

The absence of Howell-Jolly bodies or siderocytes in the peripheral blood smear is suggestive of the presence of functional splenic tissues. The CT scan can demonstrate a mass with a tissue density similar to that of the normal spleen, while magnetic resonance imaging (MRI) can demonstrate an intermediate intensity on both T1- and T2-weighted images with restricted diffusion to water [3,25]. The presence of cystic lesions is attributed to central necrosis that is due to the inadequate blood supply [26]. The diagnosis can further be confirmed using a functional imaging study which is a sensitive and noninvasive method. Technetium-99m heat-denatured erythrocytes scintigraphy is considered to be the most specific nuclear study technique that allows for the differentiation of hepatic tissue from splenic tissue. Scintigraphy with Technetium-99m sulfur colloid and indium-111-labeled platelets are considered alternative diagnostic methods but also have lower sensitivity [27,28]. Ferumoxide-enhanced MRI has been reported to detect splenic tissue as it demonstrates a brief increase in T2-signal intensity proceeded by a characteristic decrease [3]. The diagnosis of splenosis may be confirmed via these imaging modalities, thus precluding the need for invasive diagnostic procedures. However, for our patient, such imaging studies were not performed due to the acute nature of the presentation. Additionally, the associated risk of sepsis in post-splenectomy patients was considered. If nuclear studies are unavailable, a pathologic analysis of the lesions would be required for an accurate diagnosis. It should be noted, however, that the results of fine-needle aspiration might be misleading for the diagnosis of lymphoma [29].

As splenosis is a benign condition and it may have some immunologic protection against encapsulated bacteria [30], it does not require treatment in most cases. Surgical resection, either open or laparoscopic, should be performed only if required. In our case, only the nodule located in the mesoappendix was resected.

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Conclusions

Abdominal splenosis is not an uncommon condition in patients with a history of splenic injury and splenectomy. However, the involvement of the mesoappendix, which may or may not contribute to acute inflammation of the appendix, is very rare. Splenosis should be considered in the proper clinical settings and it should not distract away from the underlying etiology of acute abdomen.

Conflict of Interests

None.

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