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Case report

Perirectal intraperitoneal splenosis: A case report of MRI with laparoscopic correlation ☆☆☆

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ABSTRACT

Splenosis is a benign acquired condition, which appears after rupture of the spleen and heterotopic auto-transplantation. Mostly found as an incidental finding on cross-sectional imaging, definitive diagnosis is frequently made histologically after resection or tissue sampling. We report a case of a 36-year-old male patient who presented with increased susceptibility to infections, chronic fatigue, and a history of traumatic splenic rupture. Cross-sectional imaging showed perirectal formations within the mesorectal fascia, and extraperitoneal splenosis was suspected. Due to the radiologically unclear entity of the masses, diagnostic laparoscopy with tissue sampling was performed. Intraoperatively the masses turned out to be intraperitoneal. Histological workup showed splenic tissue, consistent with intraperitoneal splenosis after splenic rupture. In this article we want to discuss important imaging findings and their differentials, as well as clinical implications for this rare entity.

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Introduction

Splenosis is a benign condition of acquired ectopic splenic tissue, which is usually discovered incidentally and can be difficult to distinguish from malignancy in regular cross-sectional

imaging [1]. In 1910, Hermann Küttner first described a splenic tissue implantation after rupture of the spleen [2]. Two types of ectopic splenic tissue are known: First, congenital accessory spleen, which evolves from the dorsal mesogastrium. Second, splenosis, which is an acquired condition by heterotopic auto-transplantation after splenic trauma or surgery [3]. There are two hypotheses of the pathogenesis of splenosis: Intrahepatic splenosis is thought to result from a haematogenous spread through the portal vein after traumatic rupture, whereas intra- or extraperitoneal splenosis is thought to result from direct intra- or extraperitoneal spread of splenic tissue after

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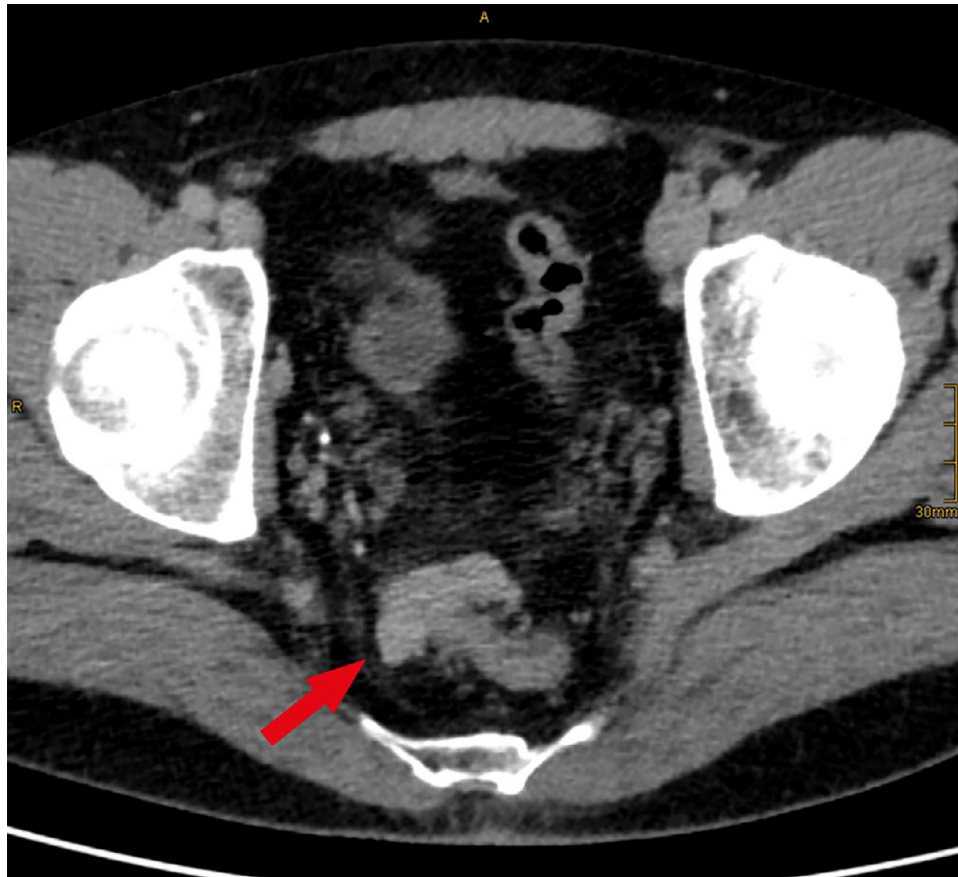


Fig. 1 – Axial contrast-enhanced CT image showed nodular perirectal masses (arrow).

trauma [4]. Those ectopic implantations retain ability to function, although their histologic pattern is different from normal splenic tissue [5,6]. In most cases, the ectopic tissue is found within the peritoneal cavity, especially on serosal surfaces of the small and large bowel [7]. Usually, splenosis remains asymptomatic, until found incidentally at screening tests, abdominal operations or autopsy. Infarction, pain, intestinal obstruction, adhesive bands and gastrointestinal haemorrhage are known complications of intraperitoneal splenosis [3]. Unless symptomatic, there is no need for treatment [3]. Different radiological examinations, such as ultrasound, x-ray, multidetector computed tomography (MDCT) or magnetic resonance imaging (MRI), can identify ectopic splenic tissue, although the differentiation from malignancy can be hard for radiologists to assess [8]. Non-invasive definitive diagnosis can be made with Tc-99m-tagged heat-damaged red blood cells scintigraphy (Tc-99m-DRBC scintigraphy), but lacking availability of Tc-99m-DRBC scintigraphy or unclear appearance on cross-sectional imaging, the diagnosis is frequently made histologically after resection or tissue sampling [9].

Case report

We report a case of a 36-year-old male patient, who presented with chronic fatigue, night sweat and increased susceptibility

to infections for years. The patient's history revealed a motor-bike accident 20 years before with splenic rupture and subsequent splenectomy. After splenectomy, the patient was only vaccinated against *Streptococcus pneumoniae*. Physical examination revealed no abnormalities and laboratory tests (including complete blood count with differential, comprehensive metabolic panel, Carcinoembryonic Antigen) were unremarkable.

To exclude malignancy, a contrast enhanced MDCT of the thorax and abdomen was done during the course. Perirectal formations with soft-tissue equivalent attenuation values were found within the mesorectal fascia (Fig. 1). No suspect lymph nodes or further tumour-suspicious masses were present. In a subsequent contrast-enhanced MRI of the pelvis, the perirectal formations showed low signal on T1- and T2-weighted images (Fig. 2A and B, 3), no diffusion restriction (not shown) and homogenous contrast enhancement (Fig. 2C and D). There was no clear connection to the intestinal lumen and on imaging, the intestinal wall was normal. From cross-sectional imaging alone, an exact diagnosis was not possible, but extraperitoneal splenosis in the mesorectum was suspected (Fig. 3).

Due to the possibility of a malignant entity, indication for a diagnostic laparoscopy with tissue sampling was made in an interdisciplinary conference. Intraoperatively, an intraperitoneal, reddish-brown, pea-sized formation next to the sigmoid colon and the small intestines was found. A second

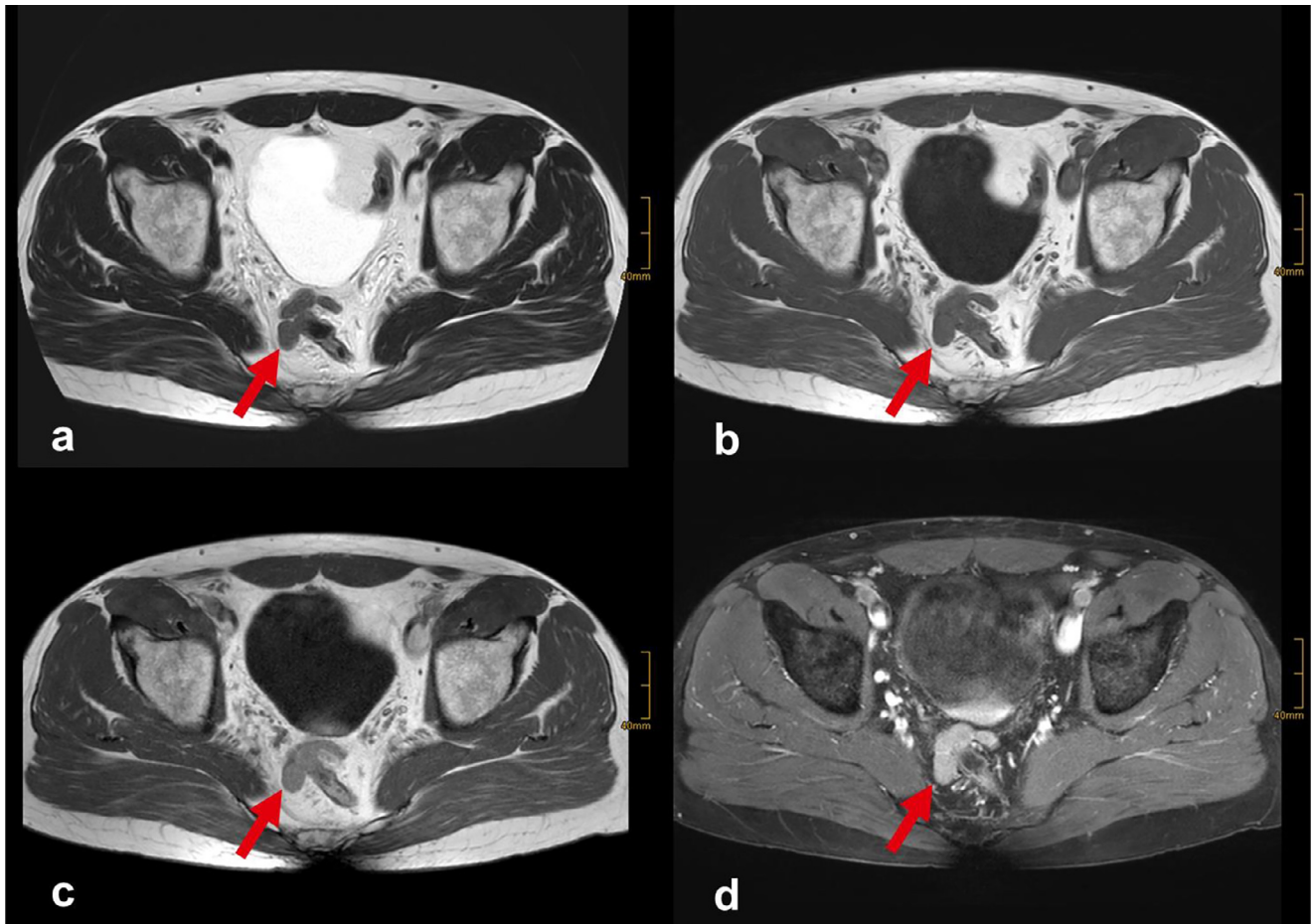


Fig. 2 – Transverse MR imaging demonstrates multiple nodular perirectal formations (arrow) with intermediate signal on plain images and homogeneous enhancement after i.v. administration of gadolinium: (A) T2-weighted and (B) T1-weighted image before as well as (C) T1-weighted without and (D) T1-weighted image with fat saturation after gadolinium administration.

intraperitoneal, reddish-brown, orange-sized formation between the anterior rectal wall and the bladder was present. After a partial resection, splenic parenchyma was histologically confirmed in a frozen section analysis. The frozen section showed normal appearing splenic tissue with trabeculae and red and white pulp, even capsular fibrous tissue, indistinguishable from tissue of a normal spleen (Fig. 4). A detachment of the splenic tissue from the rectal wall was not performed, because it appeared to be firmly attached. The rest of the splenic tissue was left untouched. Before discharge the patient received the remaining vaccinations after splenectomy.

Discussion

Splenositis is the heterotopic implantation of splenic tissue after trauma or surgery [10]. Usually it is asymptomatic, unless complications, such as infarction, pain, intestinal obstruction, adhesive bands or gastrointestinal haemorrhage, occur [7]. Finding the correct diagnosis could be difficult with common imaging methods if typical radiological features are ab-

sent. A complete radiological evaluation including CT and MRI is necessary to exclude malignancy and to determine the correct therapy [11]. Thus, it is critical to differentiate intra- and extraperitoneal lesions. Normally, a MRI examination of the pelvis is suitable to assess the location, invasiveness and exact borders of perirectal masses. On cross-sectional imaging the peritoneum is usually not visible and the exact borders and compartmentalization in the lesser pelvis have been a long-time controversy between radiologists and anatomists [12]. If a mass is found in the deep lesser pelvis on CT and MRI with close contact to the rectum and within the mesorectal fascia it is usually classified as an extraperitoneal lesion. Due to the case history and deep location of the lesions in the lesser pelvis, an extraperitoneal splenositis was suspected. In a review of the literature, we did not find a case report with extraperitoneal splenositis located in the lesser pelvis and only a few case reports of retroperitoneal splenositis [13]. Other possible locations of splenositis are intrathoracic after splenic and diaphragmatic rupture, intraperitoneal on the greater omentum, the small bowel and the diaphragm, in the pancreas, the pelvis, liver, kidney, cerebrum, and subcutaneous tissues [14]. Because of the low possibility of an extraperitoneal splenositis



Fig. 3 – Sagittal T2-weighted MR image shows the nodular formations adjacent to the rectum.

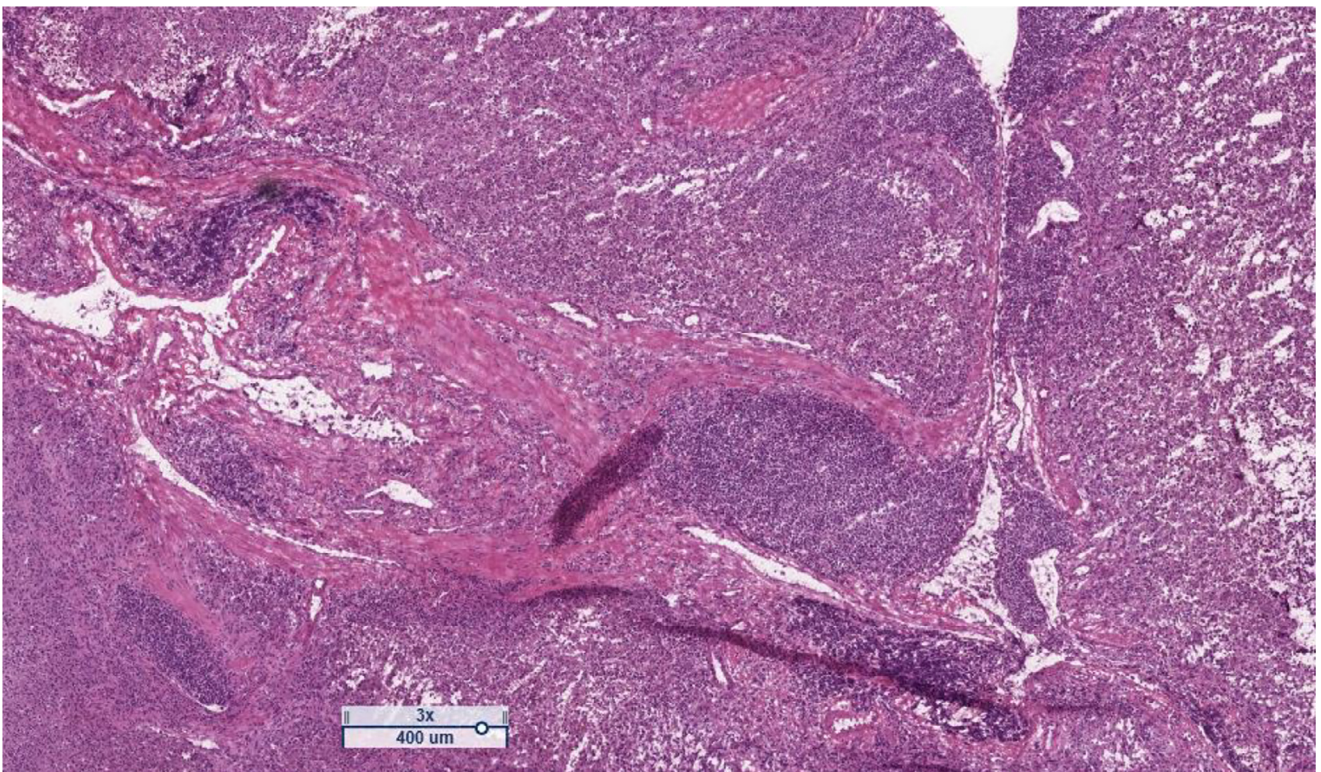


Figure 4 – Histopathological workup from the resected specimen, hematoxylin and eosin staining finally revealed normal splenic tissue.

and the possible malign differential diagnosis, such as lymphatic proliferation, sarcoma, metastases, or epidermoid cyst, decision for primary resection was made [14]. Also, no possible complications of splenosis were present. One of the most important clues to the final diagnosis in the presented case was the patient's history of a traumatic splenic rupture twenty years before. Usually, ectopic splenic tissue presents on CT and MRI with similar signals as normal splenic tissue. Principally, splenosis could have been non-invasively confirmed with a Tc99m-DRBC scintigraphy [10, 14]. On cross-sectional imaging, the deep spaces of the lesser pelvis can be difficult to differentiate and can be challenging for radiologists to assess. One explanation might be that in the CT and MRI scan the peritoneal fold reaches deeper into the rectovesical pouch and those formations are still part of the intraperitoneal cavity. Another hypothesis could be that the formations were split into intraperitoneal and extraperitoneal parts and maybe we just found the intraperitoneal formations and because the mesorectal fascia was not opened on surgery. Furthermore splenectomised patients have an increased risk to fall sick with pneumococcal pneumonia, not specified pneumonia, meningitis and septicemia [15]. Therefore patients need to receive all vaccinations according to the guidelines [16].

Conclusion

Splenosis must be considered as differential diagnosis in patients with a history of splenectomy and masses of unknown etiology on cross-sectional imaging. When presenting with symptoms relatable to a malign process and radiologically suspicious lesions in the lesser pelvis, no time to make the correct diagnosis should be wasted.

Patient consent

Written informed consent for publication was obtained from the patient.

REFERENCES

- [1] Liu Y, Ji B, Wang G, Wang Y. Abdominal multiple splenosis mimicking liver and colon tumors: a case report and review of the literature. *Int J Med Sci* 2012;9(2):174–7. doi:10.7150/ijms.3983.
- [2] Garamella JJ, Hay LJ. Autotransplantation of spleen:splenosis; case report and preliminary report of an experimental study in revascularization of the heart. *Ann Surg* 1954;140(1):107–12. doi:10.1097/00000658-195407000-00012.
- [3] Fremont RD, Rice TW. Splenosis: a review. *South Med J* 2007;100(6):589–93. doi:10.1097/SMJ.0b013e318038d1f8.
- [4] Kwok CM, Chen YT, Lin HT, Su CH, Liu YS, Chiu YC. Portal vein entrance of splenic erythrocytic progenitor cells and local hypoxia of liver, two events cause intrahepatic splenosis. *Med Hypotheses* 2006;67(6):1330–2. doi:10.1016/j.mehy.2006.04.064.
- [5] Fleming CR, Dickson ER, Harrison EG Jr. Splenosis: autotransplantation of splenic tissue. *Am J Med* 1976;61(3):414–19. doi:10.1016/0002-9343(76)90380-6.
- [6] Carr NJ, Turk EP. The histological features of splenosis. *Histopathology* 1992;21(6):549–53. doi:10.1111/j.1365-2559.1992.tb00443.x.
- [7] Akay S, Ilica AT, Battal B, Karaman B, Guvenc I. Pararectal mass: an atypical location of splenosis. *J Clin Ultrasound* 2012;40(7):443–7. doi:10.1002/jcu.20843.
- [8] Tsitouridis I, Michaelides M, Sotiriadis C, Arvaniti M. CT and MRI of intraperitoneal splenosis. *Diagn Interv Radiol* 2010;16(2):145–9. doi:10.4261/1305-3825.DIR.1855-08.1.
- [9] Short NJ, Hayes TG, Bhargava P. Intra-abdominal splenosis mimicking metastatic cancer. *Am J Med Sci* 2011;341(3):246–9. doi:10.1097/maj.0b013e318202893f.
- [10] Pichon L, Lebecque O, Mulquin N. Splenosis mimicking peritoneal carcinomatosis. *J Belg Soc Radiol* 2020;104(1):14 Published 2020. doi:10.5334/jbsr.2089.
- [11] Xuan Z, Chen J, Song P, et al. Management of intrahepatic splenosis: a case report and review of the literature. *World J Surg Oncol* 2018;16(1):119 Published 2018. doi:10.1186/s12957-018-1419-1.
- [12] Chen N, Min PQ, Liu ZY, Wu B, Yang KQ, Lu CY. Radiologic and anatomic study of the extraperitoneal space associated with the rectum. *AJR Am J Roentgenol* 2010;194(3):642–52. doi:10.2214/AJR.09.3003.
- [13] Perry KT Jr, Zisman A, Singer J, Schulam P. Splenosis presenting as a right suprarenal retroperitoneal mass. *J Urol* 2002;168(2):644–5.
- [14] Lake ST, Johnson PT, Kawamoto S, Hruban RH, Fishman EK. CT of splenosis: patterns and pitfalls. *AJR Am J Roentgenol* 2012;199(6):W686–93. doi:10.2214/AJR.11.7896.
- [15] Kristinsson SY, Gridley G, Hoover RN, Check D, Landgren O. Long-term risks after splenectomy among 8,149 cancer-free American veterans: a cohort study with up to 27 years follow-up. *Haematologica* 2014;99(2):392–8. doi:10.3324/haematol.2013.092460.
- [16] Bonanni P, Grazzini M, Niccolai G, Paolini D, Varone O, Bartoloni A, et al. Recommended vaccinations for asplenic and hyposplenic adult patients. *Hum Vaccin Immunother* 2017;13(2):359–68. doi:10.1080/21645515.2017.1264797.