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Case Report

Multimodal study of pelvic splenosis: a rare cause of abdominal pain ☆

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ABSTRACT

We present a rare case of pelvic splenosis, in a 46-year-old man, with a previous history of partial splenectomy, complaining of nonspecific pain in the lower abdominal quadrants. Splenosis is a benign acquired condition, defined as a heterotopic autotransplantation of splenic tissue in other compartments of the body, caused by rupture of the splenic capsule following trauma or splenectomy. Splenosis is often asymptomatic and incidentally found and does not require treatment. Surgery is indicated only in patients presenting with symptoms or complications. In our case, the multimodal imaging study (ultrasound, MRI, CT, and scintigraphy) allowed a correct differential diagnosis without resorting to invasive procedures, susceptible to complications

Introduction

Splenosis is a benign acquired condition defined as the heterotopic autotransplantation of splenic tissue to other compartments of the body.

The main mechanism underlying this entity is the rupture of the splenic capsule, due to trauma or surgery, allowing for direct dissemination of the splenic tissue to other sites.

Up to 67% of splenic injuries are estimated to result in splenosis [1].

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Although often asymptomatic, patients may occasionally present with nonspecific pain, secondary to infarction and/or bleeding of the implants, peritoneal adhesions and intestinal obstruction [2], and sometimes anemia due to red blood cells sequestration.

Other paraphysiological entities characterized by multiple splenic nodules include polysplenia and the presence of accessory spleens. The specific diagnosis of polysplenia syndrome is established in the presence of other findings of left-sided isomerism, without a history of splenectomy or splenic trauma. Accessory spleens, instead, are congenital, supplied by a branch of the splenic artery and are usually a few and located near the splenopancreatic or gastrosplenic ligament.

In splenosis, on the other hand, splenic nodules can be found throughout the body, and they derive their blood supply from surrounding tissues [1,3]; these represent a real diagnostic challenge because they often mimic malignant lesions.

Splenic nodules are often first seen incidentally on ultrasound or CT examinations. Neither ultrasound nor unenhanced CT can ensure the splenic origin of the masses; however, imaging features on intravenous (IV) contrast-enhanced CT can lead to the correct diagnosis. In fact, in the dynamic postcontrast study, the masses show the same density and contrast enhancement pattern as the normal splenic tissue.

Owing to the excellent soft tissue contrast resolution, MRI may be the imaging method of choice with a higher sensitivity in the diagnosis of splenosis. The advantages of MRI include the lack of ionizing radiation and the routine use of dynamic multiphasic acquisitions, which can provide higher specificity compared with single-phase CT.

Nuclear medical imaging represents an additional diagnostic tool. Either heat-damaged red blood cells tagged with 99mTc or 99mTc-nanocolloidal serum albumin can be used as radiopharmaceuticals. The former exploits the hemocatheretic action of the spleen responsible for the sequestration and destruction of senescent and damaged erythrocytes; the latter, after IV administration, is phagocyted by the reticuloendothelial system cells, particularly represented in the liver, spleen, and bone marrow [4,5]. The use of autologous platelets labeled with 111I-oxine is less common [6].

Furthermore, modern nuclear medicine techniques allow for the integration of morphological and metabolic data using SPECT/CT imaging, ensuring a better localization and characterization of the hyperaccumulation areas of the radiopharmaceutical.

Case report

A 46-year-old man presented to his GP complaining of nonspecific, severe, intermittent pain in the lower abdominal quadrants, only partially responsive to analgesics, lasting for some months. There were not any other associated symptoms. Past medical history was mute and without familiarity for neoplastic disease.

The GP prescribed an ultrasound of the lower abdomen, which revealed the presence of a gross, unevenly isohypoechoic retrovesical mass that could not be better characterized due to intestinal bloating and poor bladder filling at the time of the examination. He later underwent a MRI of the pelvis which confirmed the presence of an elongated, capsulated nodular area, with regular margins and maximum axial diameters of about 4 × 5 cm, extending longitudinally for about 6 cm. The lesion, located immediately in front of the sigmoid-rectal junction, imprinted the posterosuperior wall of the bladder in the left para-median site and compressed and dislocated the ipsilateral seminal vesicle (Fig. 1).

The lesion was homogeneously hypointense in the T1-weighted sequence, isohypointense in T2 and isointense in the STIR sequence.

In the suspicion of a heteroplastic lesion or a secondarism, the patient underwent complete laboratory investigations and instrumental diagnostic completion with contrast-enhanced CT examination.

Laboratory tests showed no abnormalities, with regular blood counts and normal inflammatory indexes, including tumor markers such as CA-125, CA 19-9, b-HCG, and alpha-fetoprotein.

On a more in-depth medical history, the patient referred he underwent partial splenectomy as a child following a splenic injury due to a car accident.

The unenhanced CT of the abdomen revealed the presence of other smaller nodular formations with the same characteristics as the pelvic one both in the subhepatic and perisplenic area (Fig. 2). All these nodules, after administration of IV contrast, showed the same enhancement pattern as the normal native splenic tissue (Fig. 3).

All the nodules had a vascular supply deriving from locoregional capillaries without a well-defined vascular hilum.

In the suspicion of multifocal splenosis with rare abdominal-pelvic localization, it was agreed not to proceed with tissue typing of these nodules by needle biopsy due to the high risk of post-procedural hemorrhage, but to perform a scintigraphic investigation in order to obtain diagnostic confirmation.

A 740 MBq dose of 99mTc-serum albumin nanocolloid was administered intravenously and a dynamic image acquisition was performed for 30 minutes after administration of the radiopharmaceutical (60 sec/frame), at the end of which static planar images in standard orthogonal projections (Fig. 4) were acquired.

The examination was then integrated with the SPECT / CT of the abdominal-pelvic area (Figs. 5 and 6) [4,5].

The scintigraphic investigation in the dynamic and static segmental acquisitions revealed 2 areas of radiocolloid hyperaccumulation located respectively in the left hypochondrium and in the left median-paramedian retrovesical side of the pelvis.

The following integration with SPECT / CT imaging confirmed the 2 abovementioned lesions and revealed the presence of two further areas of radiopharmaceutical uptake, in the left hypochondrium, close and cranial to the aforementioned one, and in the right hypochondrium, close to the VI hepatic segment. Both were less evident, and the lower uptake of these areas was explained by the interference of the physiological accumulation of the radiopharmaceutical in the spleen and liver lodge, as well as by their smaller size [7].

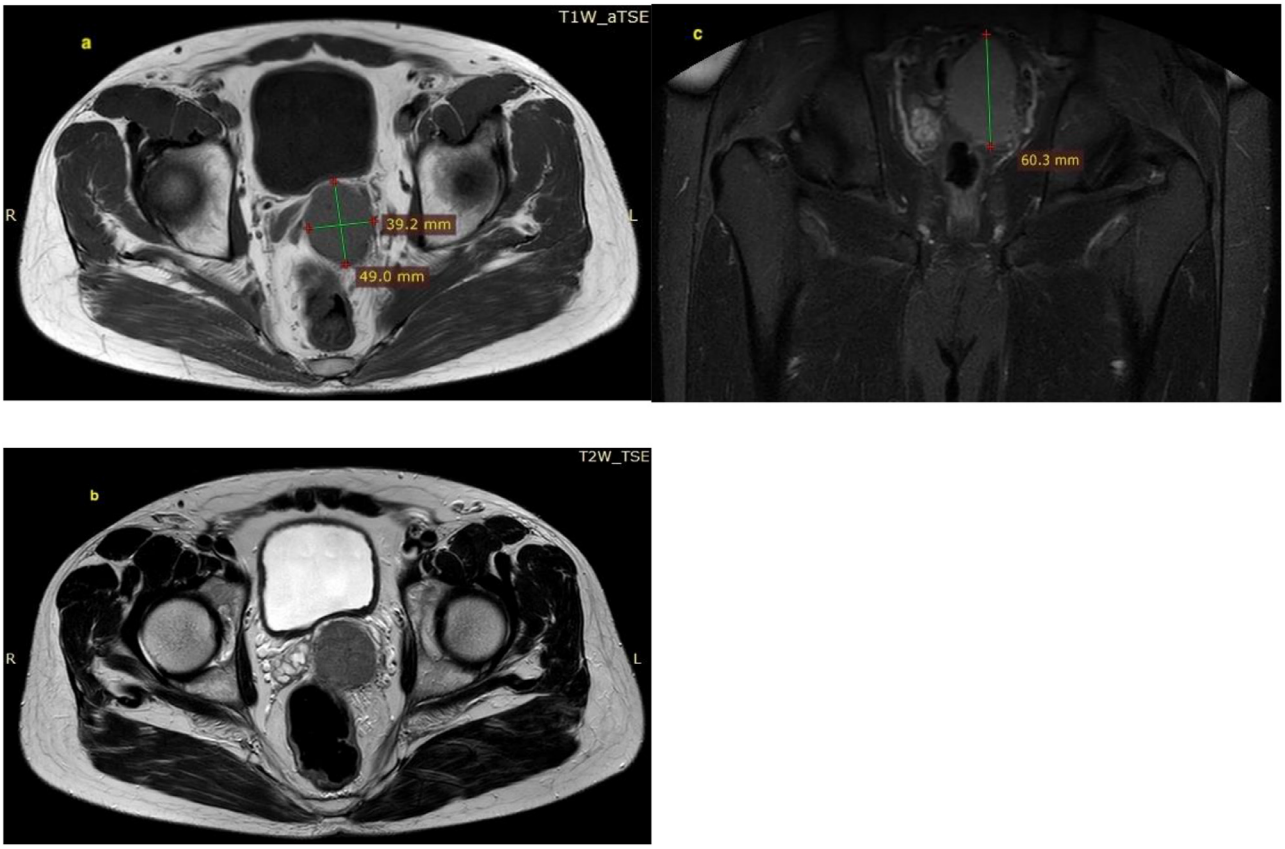

Fig. 1 – MRI study shows the presence of a pelvic mass in the retrovesical region which is homogeneously hypointense in the T1 sequence (a), isohypointense in T2 in the axial plane (b) and isointense in the coronal STIR sequence (c).

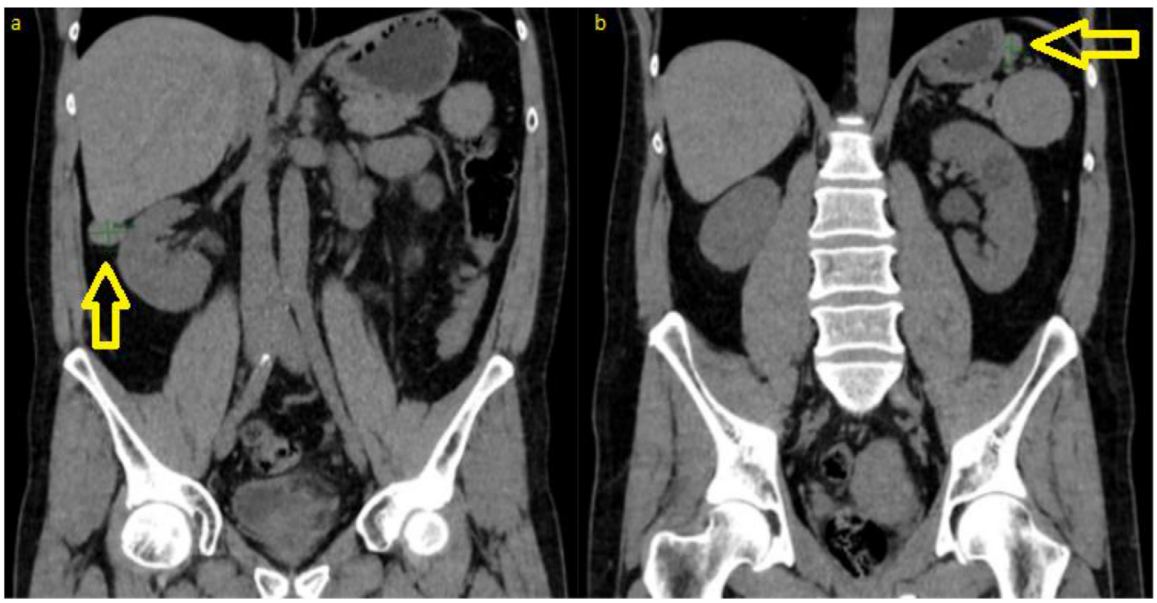

Fig. 2 – Unenhanced CT shows the presence of two nodular formations both in the hepatic area in contiguity with the VI hepatic segment (a, coronal), and in the subdiaphragmatic splenic area (b, coronal).

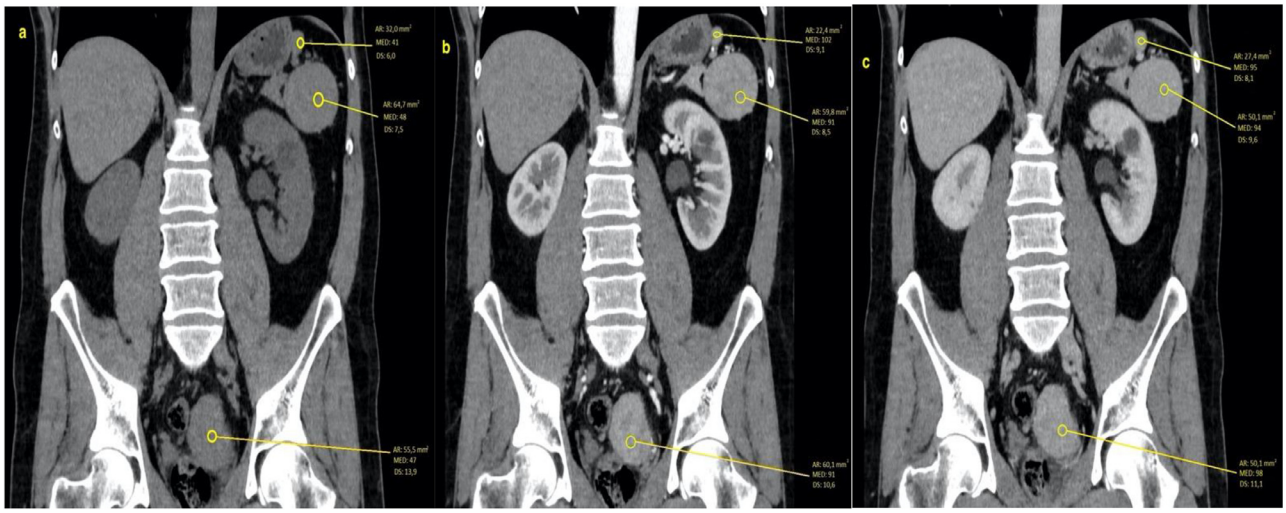

Fig. 3 – The dynamic CT study highlights how the enhancement curve of the three structures considered (native spleen, splenosis mass in the pelvic fossa and splenosis nodule in the left subdiaphragmatic site) is completely overlapping in the scans without contrast (a) in the arterial phase (b) and in the portal phase (c).

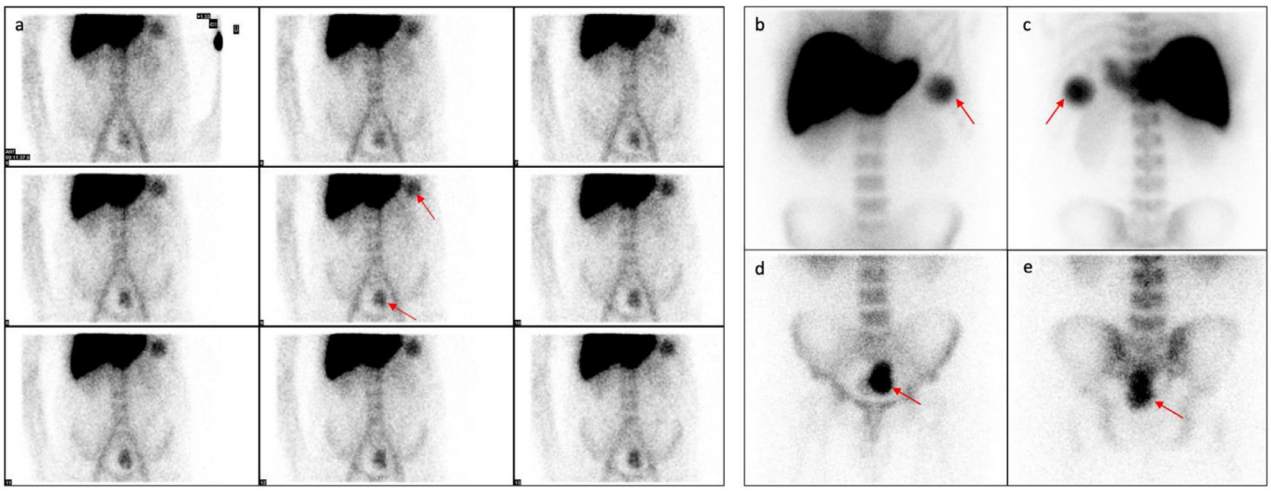

Fig. 4 – The dynamic (a) and static segmental images respectively in anterior (b; d) and posterior (c; e) projections show two areas of radiopharmaceutical hyperaccumulation in the left hypochondrium and in the pelvic area respectively (arrows).

The diagnosis of abdominal-pelvic splenosis was, therefore, confirmed and the patient was discharged due to the benignity of the condition, avoiding surgery.

DISCUSSION

Splenic trauma and splenectomy make possible the dispersion of the splenic pulp by contiguity and its implantation in the serous surface of the peritoneal cavity [2].

Splenic implants are viable tissue, nourished by adjacent vessels [8]. They can be single or multiple [9]. The degree of maturation of the implants is variable and, in some cases, they can completely replace the activity of the spleen.

Less frequently, hematogenous dissemination of splenic pulp or splenic erythropoietic progenitors may occur. This mechanism explains the finding of hepatic and intracranial splenosis foci [2].

The functional role of recurrent splenic tissue in splenosis has been studied and a certain degree of protection against bacterial infections and a lower frequency of sepsis have been highlighted [10–12].

In hematological diseases such as immune thrombocytopenia or hemolytic anemia, even following therapeutic splenectomy, the presence of splenosis or accessory spleen can lead to relapses of these pathological conditions for the same physiopathological mechanisms put in place by the native spleen [12]. After splenectomy, in fact, it is recommended to perform a follow-up CT scan to detect growing, residual, or implanted splenic tissue.

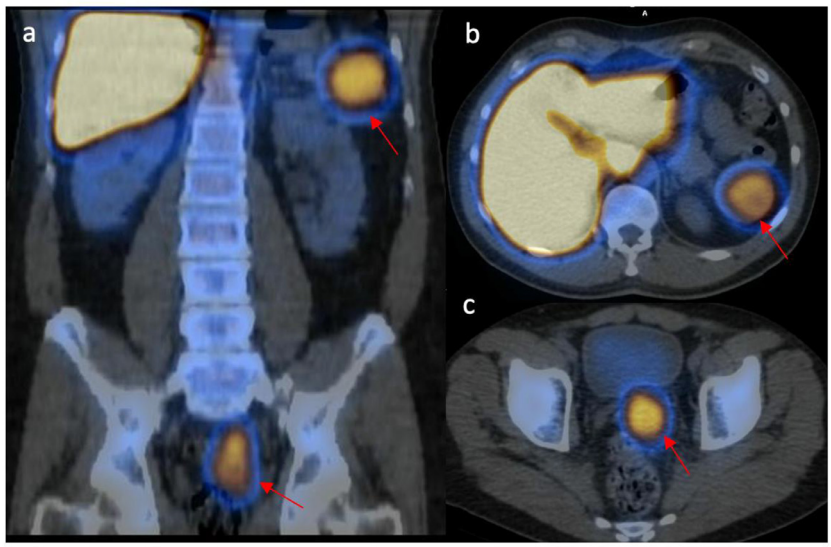

Fig. 5 – Coronal (a) and axial (b, c) projections of SPECT/CT imaging which confirm the presence of 99mTc-serum albumin nanocolloid accumulation areas in the splenic area and in the left median-paramedian retrovesical area.

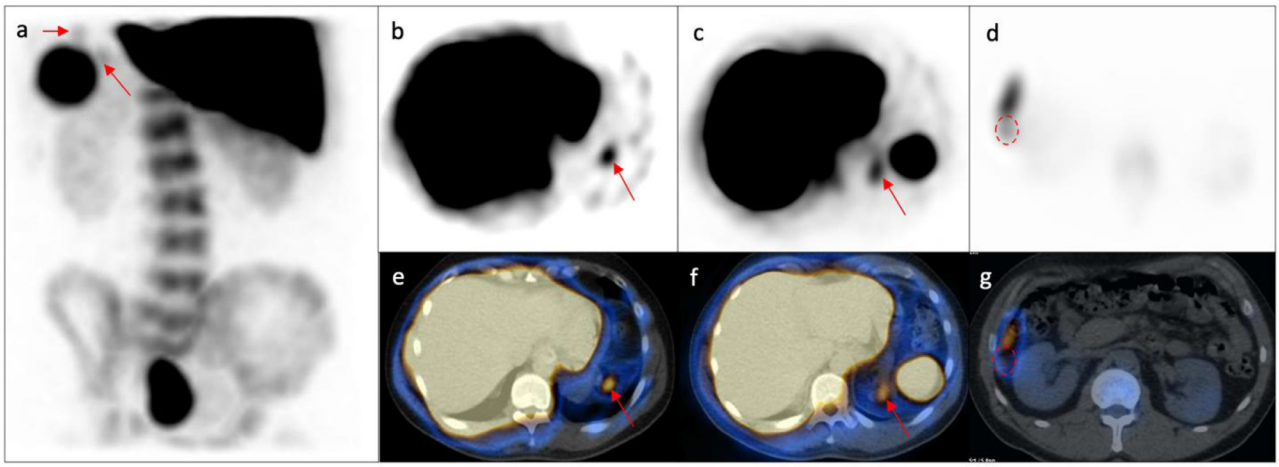

Fig. 6 – Maximum intensity projection (a) and axial projections (b, c, e, f) of SPECT/CT imaging that highlights further areas of accumulation of nanocolloids in the left hypochondrium, cranial to the area previously revealed in the splenic lodge (red arrows). Another area (d, g) is observed in the right hypochondrium, less evident due to the interference of the physiological accumulation of the radiopharmaceutical in the liver (red circle).

Splenic implants are generally smaller than 3 cm and their morphology is varied (oval, round, pedunculated). Unlike accessory spleens, which receive their blood irrigation from branches of the splenic artery, nodules of splenosis have no central artery and receive their vascularization from surrounding blood vessels that penetrate the capsule in its non-muscular portion. In most cases, a dysmorphic architecture is demonstrated due to absence of the hilum, presence of a fibrotic capsule and poor formation of germinal centers and trabecular system [13,14].

The diagnosis of this entity is generally incidental and is related to evidence of splenic implants in imaging studies or during surgical procedures [13]. Ultrasound shows nodules or round abdominal masses, with well-defined contours and solid appearance, hypoechoic in the center, with hyperechoic ridge and posterior acoustic enhancement [14]. When the location of splenosis foci allows for Doppler US, the absence of a central artery and the presence of vessels penetrating the capsule are observed, a finding that favors differentiation with accessory spleens [14].

CT allows for exact definition of the number, size, and morphology of the implants and shows their density and enhancement pattern, analogous to the splenic one [9].

MRI with IV superparamagnetic iron oxide has been used for the diagnosis of splenosis because this contrast agent is specific for the cells of the reticuloendothelial system of the liver and spleen. On unenhanced sequences, splenosis implants show homogenous hypointensity on T1-weighted im-

ages (WI) and hyperintensity on T2-weighted images (or hypointensity in case of iron deposition). Like normal spleen, splenosis shows an arterial heterogeneous/serpiginous enhancement pattern with later progressive homogenization during the dynamic studies; however, recognition of this arterial pattern may depend on the implant size, being easier to recognize in larger implants. This contrast enhancement pattern is also seen on dynamic CT imaging [15].

Scintigraphy with heat-damaged red blood cells tagged with 99mTc or 99mTc-nanocolloidal serum albumin, thanks to its high sensitivity and noninvasiveness, remains the method of choice for the differential diagnosis between malignancy and splenosis, especially in the most complex cases. A case report published by Alvarez et al [16] proved the complementary SPECT/CT investigation to be significantly advantageous to identify and localize small soft tissue lesions, such as splenic tissue residues. This evidence was also confirmed by Honger et al [17] on a greater number of patients.

However, it should be emphasized that the sensitivity of this method can be limited in case of small accessory spleens located close to the splenic hilum in non-splenectomized patients, or in proximity to areas of physiological uptake of the radiopharmaceutical [7].

Conclusion

Whichever imaging study is used (ultrasound, CT, MRI, and scintigraphy), the correct diagnosis and detailed characterization of new implants is essential since they can be confusing and mimic heteroplastic lesions, metastases, endometriosis and lymphoma. Splenosis is a benign condition and does not require treatment. Surgery is indicated only in symptomatic or complicated patients, in recurrent post-splenectomy hematological disease, or when the diagnosis is uncertain.

Patient consent statement

Informed written consent was obtained from the patient for publication of the Case Report and all imaging studies. Consent form on record.

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