Journal of Cardiovascular Disease Research

ISSN: 0975-3583,0976-2833 VOL15, ISSUE 01, 2024

HEPATIC SPLENOSIS- A DIAGNOSTIC DILEMMA WITH REVIEW OF LITERATURE

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Received Date: 20/12/2023 ***Acceptance Date: 06/01/2024***

Abstract

Splenosis is ectopic splenic tissue caused due to autotransplantation of one or more focal deposits of splenic tissue in various compartment of body.Hepatic splenosis is rare cause of focal liver lesion indistinguishable from primary and metastatic liver tumours . We are presenting a case report of 45 years old male detected a focal liver lesion on CT and MRI imaging ,had a history of splenectomy for splenic trauma. In view of suspicion of liver neoplasm further laproscopic resection of focal liver lesion was planned. Histopathological examination showed hepatic splenosis. With this case we would like to highlight the importance of clinical and diagnostic suspicion of intrahepatic splenosis in patient with history of splenectomy / splenic rupture. Further following diagnostic crietria[1] (as per the review of literature). 1.history of splenic trauma or splenectomy; 2. lesion(s) with a surrounding rim, particularly near the liver capsule identified by CT scanning; 3 .findings on superparamagnetic iron oxide-enhanced magnetic resonance imaging or technetium-99m heat-damaged red cell scanning; and 4.histopathological findings (needle biopsy or surgical pathology). The following diagnostic process is also proposed: suspect diagnosis when criteria 1 and 2 are met; make diagnosis when criterion 3 is met; confirm diagnosis when criterion 4 is met. Laparotomy is recommended for either diagnosis or treatment when invasive procedures are necessary.

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Introduction -

Splenosis is a benign condition caused due to autotransplantation of one or more focal deposits of splenic tissue in various compartment of body.Hepatic splenosis is rare cause of focal liver lesion indistinguishable from primary and metastatic liver tumours , commonly resulting from traumatic rupture or splenectomy [1] .Hepatic splenosis is rare and usually diagnosed incidentally. Due to its low prevalence, it is difficult to diagnose by non-invasive methods, particularly when the mass presents as malignant disease on imaging or the patient has a risk for hepatic malignancy. Hence, the diagnosis of hepatic splenosis may require further investigation

Case presentation

A 45 year old male presented with right upper quadrant discomfort and occasional pain of long duration . He had no history of weight loss, abdominal pain, or jaundice. . His past medical history revealed history of splenectomy for splenic trauma . He is not an alcoholic/smoker. There was no positive sign on physical examination, except for a previous surgical scar. His liver function was normal and graded as A (score 5)according to the Child–Turcotte–Pugh classification. His α fetoprotein (AFP) level ,carcinoembryonic antigen and carbohydrate antigen were normal range . Chest radiography was normal. Ultrasonography showed a well-demarcated hypoechoic lesion in right lobe segment VI , appears subcapsular . Contrast-enhanced CT abdomen revealed 2.6 x1.8cm isodense lesion with hypoenhancement on arterial and portal phase with no significant washout on delayed phase.No evidence of liver parenchymal disease. In view of inconclusive CT finding ,MRI liver with contrast was done ,lesion appears hypointense on T1 and hyperintense T2 with restricted diffusion and shows mild enhancement on arterial phase with mild washout on portal venous and delayed phase imaging . With inconclusive MRI and CT finding futher laproscopic resection of liver lesion was planned.

Further histopathological section shows well circumscribed lesion composed of predominantly thin wall vascular sinusoids separated by lymphoid collections. The vascular spaces are consistent with inconspicuous endothelial lining. Some of the lymphoid follicles had eccentrically placed arterioles . Negative for overt atypia in the endothelial cells or stromal lymphoid collections . Histological feature were consistent with hepatic splenosis.Adjacent hepatic parenchyma including the biopsy shows no diagnostic histologic abnormalities .

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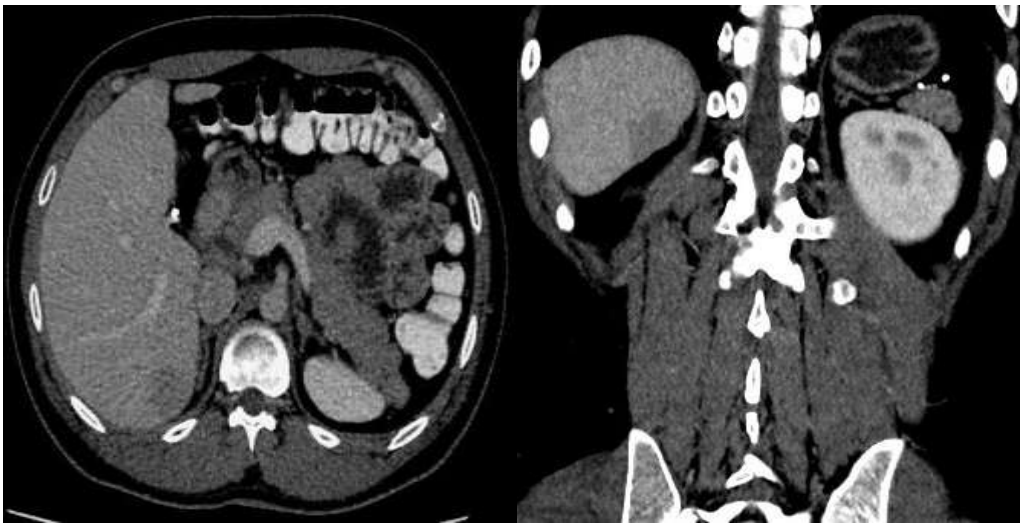

Fig.1 The axial contrast CT section shows well-demarcated subcapsular isodense lesion in right lobe segment VI with hypoenhancement on arterial and portal phase with no significant washout on delayed phase.

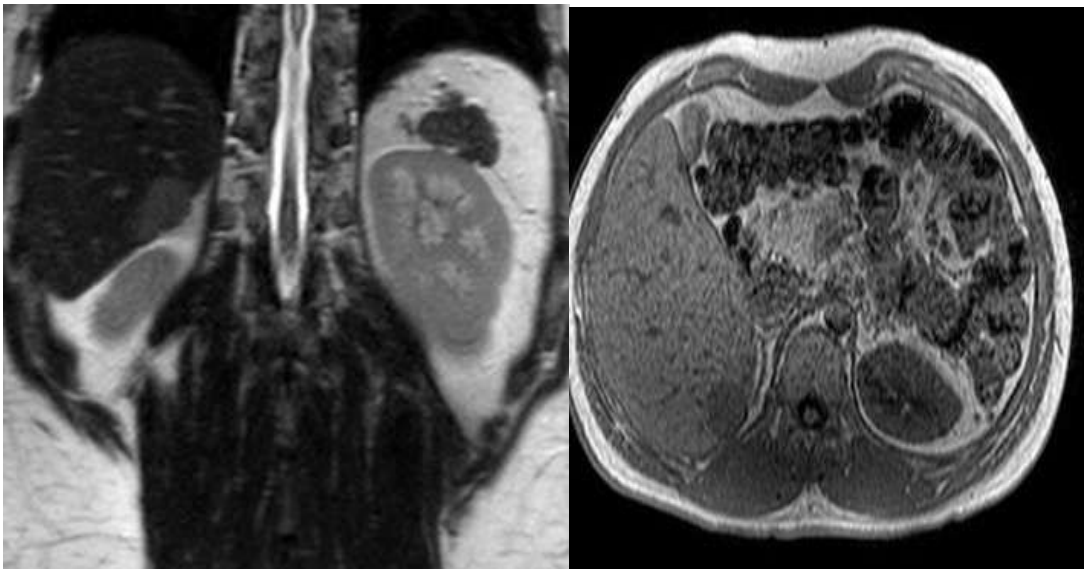

Fig.2 MRI T1 and T2 weighted images shows welldemarcated T1 hypointense and T2 hyperintense peripheral subcapsular lesion .

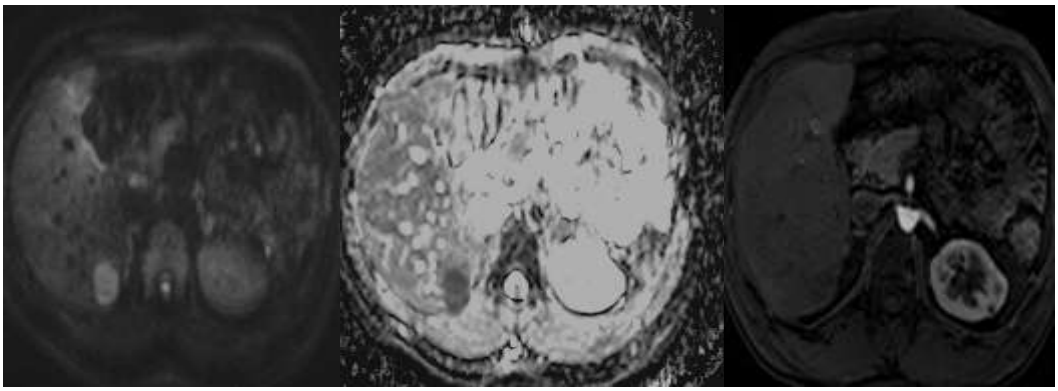

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Fig 3- Lesion appear hyperintense in DWI with signal drop in ADC, showing diffusion restriction. Post contrast arterial phase lesion shows mild enhancement .

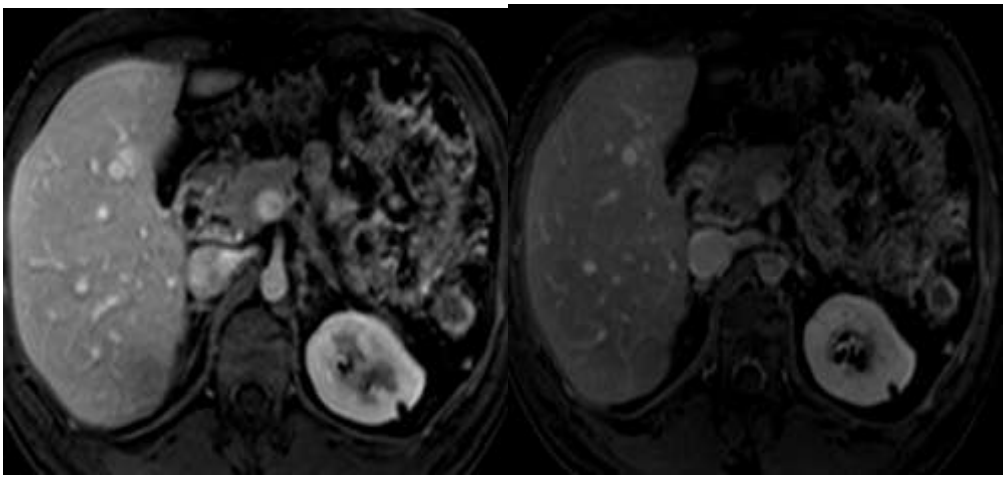

Fig 4- MRI postcontrast venous and delayed phase imaging shows mild washout

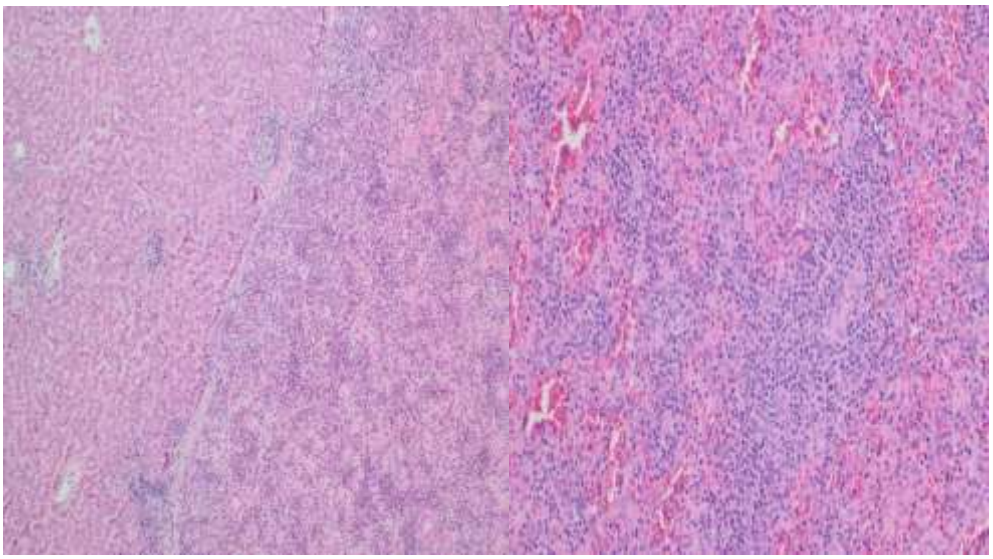

Fig 5 - Hematoxyline and eosin stains- shows splenosis with lymphoid follicular aggregates and sinusoidal strcutures. A capsule clearly separates liver and spleen parenchyma.

Discussion

The term "splenosis" was first used in 1939 by Buchbinder to describe a case in a young woman thought to have endometriosis [2] . Splenosis is defined as the autotransplantation of splenic tissue, usually occurring months to years following abdominal trauma involving rupture of the splenic capsule. CT imaging shows 1 or more homogenous, solid, and non-calcified nodules. A diagnosis can be made by demonstrating uptake of 99m-Tc sulphur colloid or 99m-Tc-labeled heat-denatured erythrocytes within the splenules, although the latter has higher specificity . Incidence of splenosis after splenic rupture as high as 76%.The splenic tissue can implant in

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various locations. These include, in descending order of frequency: the serosal surface of the small intestine, greater omentum, parietal peritoneum, Surface of the large intestine , mesentry , undersurface of diaphragm and the thorax . As in this case, however, intrahepatic splenosis is rare, with only a handful of cases found in the literature.

Sites of splenosis

1. Thorax: mostly occurs with a simultaneous diaphragmatic and splenic rupture [3].Splenic tissue is then transported to the left hemithorax, parietal, or visceral pleura.

2. Abdomen: most frequent locations are greater omentum,small bowel serosa, parietal peritoneum, under surface of the diaphragm [4]. These implants can be confused with primary or metastatic malignancy or endometriosis.

3. Intrapancreatic-accessory spleen or splenosis: very rare. In pre-contrast and post-contrast-enhanced CT and MRI images, intrapancreatic accessory spleen show similar characteristics to the orthotopic spleen. CT and MRI used in combination with DWI are important in the diagnosis [5].

4. Pelvis: can present as pelvic nodules. Can mimic metastasis, endometriosis, ovarian, uterine, and cervical masses.

5. Rare sites: liver, kidney, cerebrum, and subcutaneous tissues.

Intrahepatic splenosis is very rare as in our case, occur via the invagination of splenic implants or via splenic vein emboli. These explain their frequently subcapsular location. It can be confused with hepatic adenoma, hepatocellular carcinoma, hemangioma,lymphoma or metastasis [5,6].

Ectopic splenic tissue in the abdominal cavity is present in more than 60% of patients after traumatic splenic rupture;however, isolated hepatic localization is described only individual cases.

Splenosis is mostly seen in young males, due to the higher incidence of splenic injury within this group. The latency period between splenic injury and detection of splenosis is between 5 and 10 years in most cases.

Splenosis is notorious for mimicking benign or malignant neoplastic processes that can lead to a very expensive work-up. Generally, the splenic implants are numerous and are located within the peritoneal cavity; however, extra-abdominal splenosis does occur. The implants are rarely clinically significant and are incidental findings at autopsy or at abdominal operation, unrelated to and distant from the splenic trauma and splenectomy. Splenic implants retain their ability to function, and recurrence of a hematologic disease for which the spleen was previously removed should alert the clinician to the possibility of splenosis. The differential diagnosis includes accessory spleens, endometriosis, hemangiomas and metastatic cancer [7].

In this case, the patient had a history of splenectomy at age 30 and was unaware of any previous work-up for remnant splenic tissue.

Magnetic resonance imaging (MRI) characteristics suggesting this diagnosis are described. The lesions were mainly hypointense on T1- and hyperintense on T2-weighted images. Normal splenic tissue has the most restricted diffusion with the lowest ADC values as compared to normal intraabdominal organs. After administration of small iron oxide particles (SPIO-

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Endorem), the lesions remained slightly hyperintense relative to the hypointense liver parenchyma but showed a 50% loss in signal intensity [8]. MRI with super paramagnetic iron oxide (SPIO) has been used for the diagnosis of splenosis as this contrast agent is specific for cells of the reticuloendothelial system in the liver and spleen.As reported, intrahepatic splenosis will remain hyperintense relative to the liver parenchyma, while hepatocellular carcinoma (HCC) will become hypointense after the SPIO administration. Intrahepatic splenosis can be confused with HCC, adenoma, or other liver diseases, leading to unnecessary surgery or other invasive treatments. Therefore, more sensitive novel methods to diagnose intrahepatic splenosis are needed.

Role of nuclear scintigraphy: The splenic tissue in splenosis does not show Howell jolly bodies, Heinz bodies, and other erythrocyte abnormalities in the peripheral smears of asplenic patients with splenosis. Nuclear scintigraphy done using heat-damaged red blood cells tagged with technetium 99 (Tc99) is currently the diagnostic tool of choice due to the high uptake of damaged erythrocytes by the splenic tissue [9].

Hence, scintigraphy with Tc99-labeled heat damaged RBC is preferred as it is more sensitive and specific than Tc99- labeled sulfur colloid scintigraphy and can noninvasively confirm the diagnosis of splenosis [10].

Further following diagnostic crietria[1] (as per the review of literature)is proposed .

1.history of splenic trauma or splenectomy; 2. lesion(s) with a surrounding rim, particularly near the liver capsule identified by CT scanning; 3 .findings on superparamagnetic iron oxide-enhanced magnetic resonance imaging or technetium-99m heat-damaged red cell scanning; and 4.histopathological findings (needle biopsy or surgical pathology). The following diagnostic process is also proposed: suspect diagnosis when criteria 1 and 2 are met; make diagnosis when criterion 3 is met; confirm diagnosis when criterion 4 is met. Laparotomy is recommended for either diagnosis or treatment when invasive procedures are necessary.

Conclusion

Splenosis is a benign entity, with the greatest importance being the need to distinguish them from more sinister pathology. The importance of suspicion for this diagnosis should be highlighted when tumor-like lesions disclosed on imaging occurs in a patient with a splenic injury or removal in the past. (99m)Tc labelled heat-denatured erythrocyte scintigraphy can be helpful to the diagnosis since it may avoid the performance of biopsies or surgical resections [11].

Intrahepatic splenosis may be beneficial in patients who have undergone splenectomy because it can replace part of immunologic function of the removed spleen .Therefore, conservative treatment is strongly recommended for asymptomatic intrahepatic splenosis except for some special situations [13] .

Consent- This Gentleman has kindly given informed consent for publishing this case report.

Competing interests- The authors declare that they have no competing interests.

ISSN: 0975-3583,0976-2833 VOL15, ISSUE 01, 2024

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