



# Post-traumatic multifocal abdominal splenosis.

## The role of the clinical history.

### Case report and review of literature



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#### Post-traumatic multifocal abdominal splenosis. The role of the clinical history. Case report and review of literature

**BACKGROUND:** Splenosis is a benign clinical condition caused by the heterotopic autotransplantation of spleen's tissue typically occurring after spleen rupture. Splenosis may be asymptomatic and found accidentally. When signs and symptoms occur they are due to mass effect or bleeding of the splenic nodules.

**CASE REPORT:** 74-years-old male presenting with intestinal sub-occlusion and past medical history of post-traumatic splenectomy at 18-years-old. Based on TC findings of multiple hyperenhanced solid lesions located in greater omentum, mesentery and parietal peritoneum of right pelvic walls, the presumptive diagnosis was peritoneal carcinomatosis of unknown primary site. Stenosis of a ileum loop in the right pelvis, with dilatation and faecal stasis of the upstream loops proximal, required surgical procedure. At the opening of the peritoneal cavity the multifocal lesions varied in size, were reddish blu color, sessile, lobulate and with strong adhesions to the visceral peritoneum. Omentectomy and the blunt excision of 3 extraparietal solid nodules, which had tenacious adhesions with stenotic ileum loop serosa for the lenght of 8 cm, were performed. Histopathological examination of surgical specimens showed splenic tissue with red pulp.

**CONCLUSION:** CT scan usually do not allow to make a certain diagnosis of splenosis, so the clinical history of splenic trauma or splenectomy, positive in all cases reported in literature, represent the key in the diagnostic pathway of splenosis. Management should be conservative as much as possible nonetheless in abdominal splenosis the surgical approach should be chosen for the symptomatic patients who present abdominal pain, occlusion or bleeding.

**KEY WORDS:** Abdominal, Splenosis, Spleen, Surgery

#### Introduction

Splenosis, defined as the presence of ectopic splenic tissue, is a rare and benign clinical condition caused by

the heterotopic autotransplantation of spleen's tissue usually occurring after abdominal trauma with spleen rupture.

Heterotopic spleen tissue has been described in thorax, peritoneum, retroperitoneum and pelvic cavity and the differential diagnosis with neoplasm can be challenging<sup>1,2</sup>.

Several cases of splenosis have been described in literature. Thoracic splenosis is reported after penetrating thoraco-abdominal trauma, in this case sclerosing hemangioma or low grade lymphoma were suspected at first<sup>3</sup>. Hepatic splenosis, with nodules presented as hepatocel-

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lular carcinoma, has been reported as well <sup>4</sup>. A rare case of duodenal splenosis simulating a gastro-intestinal-stomach-tumor was published by Ylei et al in 2014 <sup>5</sup>.

In the majority of cases the patients are asymptomatic or paucisymptomatic and splenic nodules are found accidentally <sup>6</sup>.

When signs and symptoms occur, they are usually due to mass effect of the splenic nodules. In the abdominal cavity, the spleen tissue growth in the intestinal wall thickness, can lead to bleeding or mechanical bowel occlusion <sup>7</sup>.

Although the histopathological examination can be considered the gold standard to confirm the diagnosis of splenosis, this particular clinical condition should be taken into consideration in all patients presenting with a positive clinical history of post-traumatic splenectomy, in order to avoid unnecessary surgery <sup>4</sup>.

## Case Report

We present a case of 74 years old Caucasian male in Hemodialytic treatment from October 2018 for chronic kidney disease in Diabetic nephropathy with past medical history positive for post-traumatic splenectomy at 18 years old, who presented to our attention for dyspeptic syndrome, Cholelithiasis and Intestinal Sub-Occlusion. Abdominal contrast-enhanced CT showed: presence of multiple hyperenhanced solid lesions located in greater omentum, mesentery and parietal peritoneum of right pelvic walls; surface irregularity and multiple hypodense lesions of the liver, of which the largest in the VII segment with a diameter of 26 mm, mimics metastases; cholelithiasis, and stenosis of a ileum loop in the

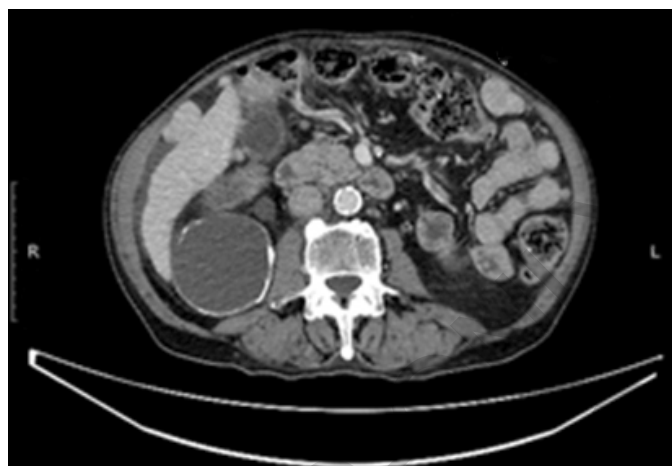


Fig. 1: Contrast-enhanced spiral CT. Axial section. 3 cm low density nodule in the sub capsular liver parenchyma and ascitic fluid.



Fig. 2: Contrast-enhanced spiral CT. Axial section. Multiple solid hypodense lesions in the greater omentum (the biggest shows 2.5 cm of maximum diameter) with perifocal edema.

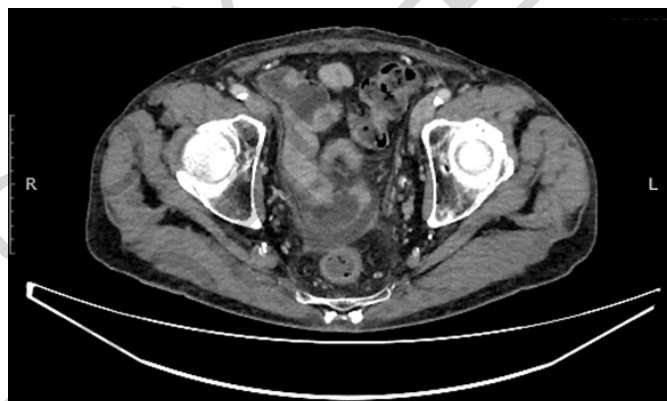


Fig. 3: Contrast-enhanced spiral CT. Axial section. Solid hyperdense nodule in the left pelvis.



Fig. 4: Contrast-enhanced spiral CT. Coronal section. Multiple hypodense solid lesions adhering to the visceral peritoneum.



Fig. 5: Surgical findings. Multiple splenic implants causing ileal loop stenosis.

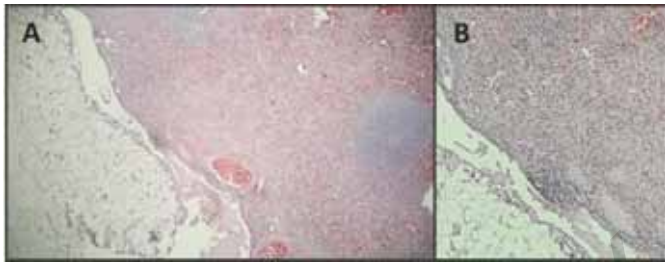


Fig. 6: Pathologic findings. Hematoxylin and eosin stained; original magnification A x4 and B x10. The appearance of the surgical sample showing splenic tissue with red pulp and with pulp near adipose tissue lined by monolayer of low cuboidal mesothelial cells.

right pelvis with dilatation and faecal stasis of the upstream loops proximal; free intra abdominal fluid was found in the perihepatic space and in Douglas pouch (Fig. 1-4).

Based on radiological findings the presumptive diagnosis was peritoneal carcinomatosis of unknown primary site.

Tumor markers were mildly positive for Alpha-fetoprotein (AFP 178 ng/ml nv <9) and Carcinoembryonic antigen (CEA 5.0 ng/ml nv <5). Carbohydrate antigen 19-9 (CA19-9 17 U/ml, nv <37) was negative.

Taken into account the clinical condition of small bowel occlusion and the ileum loop stenosis at the radiological images the surgical procedure was the treatment of choice.

At the opening of the peritoneal cavity through midline laparotomy, multifocal lesions in the greater omentum and in the mesentery were identified. The lesions varied in size, were reddish blue color, sessile, lobulate and with strong adhesions to the visceral peritoneum (Fig. 5).

The ileum loop stenosis and the upstream loops dilatation were due to extraperietal abnormal tissue implantations for the length of 8 cm.

We performed the Omentectomy and the blunt excision of 3 solid nodules which had tenacious adhesions with stenotic ileum loop serosa.

The surgical specimens underwent to histopathological examination, showing splenic tissue with red pulp and with pulp near adipose tissue lined by monolayer of low cuboidal mesothelial cells (Fig. 6).

## Discussion

This case confirms the difficulties to diagnose splenosis during clinical practices. Despite the current noninvasive imaging technologies, splenosis often mislead to a diagnosis of metastatic cancer <sup>8</sup>.

In our patient, in accordance with the current literature, the multiple splenic implantations were found incidentally at the contrast enhanced CT scan <sup>9</sup>. The main aim, not possible in all cases, should be to diagnose splenosis with the most modern non invasive imaging technologies in order to avoid unnecessary explorative laparotomies <sup>8</sup>. Unfortunately MRI and CT scan usually do not allow to make a certain diagnosis of splenosis <sup>9</sup>.

In light of the limits of both CT scan and MRI, a clinical history of splenic trauma or splenectomy, positive in all cases reported in literature, represent the key in the diagnostic pathway of splenosis <sup>6,10</sup>.

Although we were aware of the past splenectomy after splenic trauma in our patient, we underestimated this anamnestic data which could be critical to put abdominal splenosis in the differential diagnosis with metastatic malignancy.

The correct management, both diagnostic and therapeutic of this rare clinical entity should be conservative as much as possible nonetheless in abdominal splenosis the surgical approach, laparoscopic when possible, should be chosen for the symptomatic patients who present abdominal pain, occlusion or bleeding <sup>11,12</sup>.

When surgical approach, open or laparoscopic, is chosen also without any symptom the surgeon should perform a simple biopsies without proceed to the excision of all the splenic implantations. The functional heterotopic splenic tissue improves the immunosuppression typical in asplenia condition <sup>13</sup>.

## Conclusion

In conclusion we identified four critical points which lead us to miss the diagnosis: 1) the rareness of post-traumatic abdominal splenosis in the clinical practice, 2) the underestimation of a positive past clinical history for splenic trauma, 3) the lack of proportionality between the value of the positive tumor markers and a metastatic malignant disease, 4) the same contrast enhancement features between the implantations and the spleen was missed.

## Riassunto

**INTRODUZIONE:** La splenosi è una condizione clinica benigna di non comune riscontro. È caratterizzata dall'autotrapianto eterotopico di tessuto splenico dopo trauma addominale con rottura della milza. Nella maggior parte dei casi i pazienti sono asintomatici o paucisintomatici e i noduli splenici sono reperti accidentali. Più raramente, a causa dell'accrescimento intramurale o della compressione estrinseca, i noduli splenici possono essere causa di sanguinamento o di ostruzione intestinale. Essi costituiscono un vero dilemma diagnostico poiché simulano una lesione neoplastica metastatica e solo l'esame istopatologico può confermare la diagnosi di splenosi addominale.

**CASO CLINICO:** Uomo di 74 anni, con anamnesi positiva per splenectomia post-traumatica all'età di 18 anni, si presenta alla nostra attenzione per subocclusione intestinale. La TC addominale con mdc mostra la presenza di multiple lesioni solide con intenso enhancement post-contrastografico in sede sottodiaframmatica. Le lesioni appaiono localizzate nel fegato, nel grembiule omentale, sul mesentere dell'intestino tenue e nel peritoneo parietale dell'emiscavo pelvico destro; si associa evidenza di stenosi di un'ansa ileale dello scavo pelvico con dilatazione e stasi fecale sovra-stenotica. Sulla base dei risultati radiologici si avanza sospetto diagnostico di carcinomatosi peritoneale da tumore primitivo a sede occulta.

Tenuto conto del reperto TC di stenosi di ansa ileale, in paziente con quadro clinico di subocclusione intestinale, la procedura chirurgica è stata ritenuta il trattamento di scelta. All'apertura della cavità peritoneale si evidenziavano nel fegato, grande omento e mesentere lesioni multifocali di diametro variabile, colore blu rossastro, sessili, lobulate, di consistenza parenchimatosa e tenacemente adese al peritoneo viscerale. La stenosi dell'ansa ileale, causa della dilatazione sovrastenotica, era determinata da un impianto extraparietale delle suddette lesioni della lunghezza di circa 8 cm. È stata eseguita l'omentectomia e sono stati rimossi, con manovra delicata, tre gettoni solidi tenacemente adesi alla sierosa dell'intestino tenue substenotico. L'esame istopatologico dei campioni chirurgici mostrava trattarsi di tessuto splenico con polpa rossa.

**CONCLUSIONE:** Nonostante la disponibilità delle tecnologie di imaging, la splenosi resta di difficile identificazione strumentale, inducendo erroneamente alla diagnosi di una malattia neoplastica metastatica da tumore primitivo occulto. Alla luce dei limiti diagnostici della TC, una storia clinica di trauma splenico o splenectomia, positiva in tutti i casi riportati in letteratura, rappresenta l'elemento più importante per avanzare il sospetto di splenosi addominale. La corretta gestione diagnostica e terapeutica di questa rara entità clinica dovrebbe essere il più

possibile conservativa. Nei casi di splenosi addominale l'approccio chirurgico, open o laparoscopico, va riservato ai soli pazienti sintomatici per dolore addominale, subocclusione intestinale e sanguinamento, limitandosi quando possibile all'esecuzione di un semplice prelievo biotico della lesione necessario per una diagnosi di natura. Si consiglia di non procedere alla rimozione di tutti gli impianti splenici che compensano, almeno in parte, l'immunodepressione tipica dell'asplenia.

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