

## Case Report

### THORACO-PARAPERICARDIAL SPLENOSIS: FEATURES OF A RARE DIAGNOSIS AND A CASE REPORT

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#### Summary

*Introduction:* Thoracic splenosis is an acquired rare condition, resulting from heterotopic autotransplantation of splenic parenchyma into chest, usually after traumatic spleen and diaphragm rupture.

*Methods and Results:* This paper describes the thoracic splenosis, as an incidental diagnosis and asymptomatic condition, 34 years after a thoracoabdominal trauma in a patient who underwent radiological investigations due to a 15 kg weight loss in the previous four months: the only abnormalities found were thoraco-parapericardial and abdominal nodules of splenosis.

*Discussion:* Thoracic splenosis is a rare condition, usually following a thoracoabdominal trauma with concomitant lesions of the spleen and diaphragm, which allow thoracic implantation of splenic tissue. Patients are generally asymptomatic, and the condition is discovered incidentally during radiological investigations, which allow certain diagnosis without using invasive methods. Excision of splenosis should be considered for symptomatic patients or unconfirmed diagnosis.

*Conclusions:* Thoracic splenosis is a rare condition. Imaging investigations allow to diagnose the condition, and excision of splenosis is not suggested for asymptomatic patients.

#### Introduction

Splenosis is a rare and benign acquired condition caused by the heterotopic autotransplantation of focal deposits of splenic tissue to an abnormal location resulting in total or partial splenectomy, often but not only, traumatic occurrence that causes a splenic rupture. As a rare condition, thoracic splenosis may be found in 18% of patients after splenic rupture (1,2). The symptoms of splenosis are infrequent and the condition is discovered incidentally. Patient history, radiological investigations and nuclear imaging may allow certain diagnosis without invasive procedures (3).

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### Methods and Results

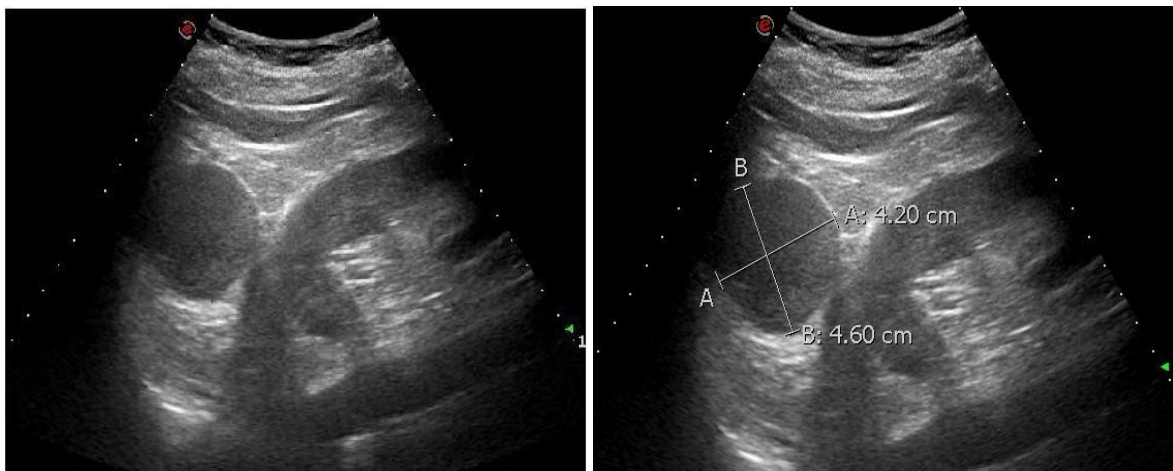
The purpose of this paper is to describe the thoracic splenosis, as incidental diagnosis and asymptomatic condition, diagnosed in a patient 34 years after thoracic and abdominal trauma.

We present the case of a 55-year-old male, born in 1964, who arrived to our department in order to undergo a chest x-ray and abdominal ultrasound due to a 15 kg weight loss in the previous four

months. In his history, he reported of a severe trauma in 1982 involving multiple rib fractures, a left side diaphragmatic tear, a splenic tear and post-traumatic splenectomy. While the chest x-ray was interpreted as normal (**Figure 1**), the abdominal ultrasound showed a round homogeneous nodule (46 x 42 millimeters in diameter) in the upper left abdominal cavity and it was interpreted as possible splenosis (**Figure 2a and Figure 2b**).



**Figure 1.** Thoracic X-rays.



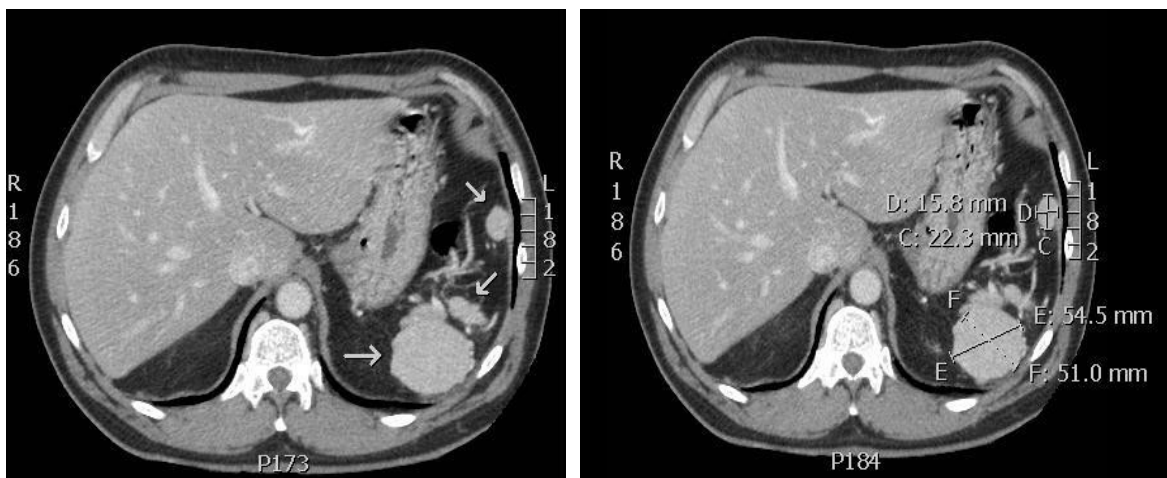
**Figure 2a (left) —2b (right)** . US: round and homogeneous nodule in upper left abdominal cavity.

Thus, an abdominopelvic CT was performed to confirm the diagnostic hypothesis. In the upper left abdominal cavity the CT showed few round, homogenous and hyperdense soft tissue nodules (25 to 75 HU), well-circumscribed, with a maximum size of 54,5 x 51 millimeters (Figure 3) and all of them demonstrated CT contrast enhancement like the spleen at portal

phase, showing attenuation values ranging from 65 to 150 HU (Figure 4a and Figure 4b). Furthermore, in sections above the diaphragm, we found two other nodules in the fat tissue of the parapericardial space, (maximum size: 18 x 14,5 mm), with the same CT features of the nodules localized in the left abdominal cavity (Figure 5a, Figure 5b and Figure 5c). These thoraco-



**Figure 3.** CT before administration of intravenous material contrast: homogenous, hyperdense and well-circumscribed soft tissue nodules, in upper left abdominal cavity.



**Figure 4a (left) – 4b (right).** CT after administration of intravenous material contrast (equilibrium phase).

parapericardial and abdominal nodules were the only abnormalities we found. The history and CT features allowed us to diagnose thoracic (and abdominal) splenosis.

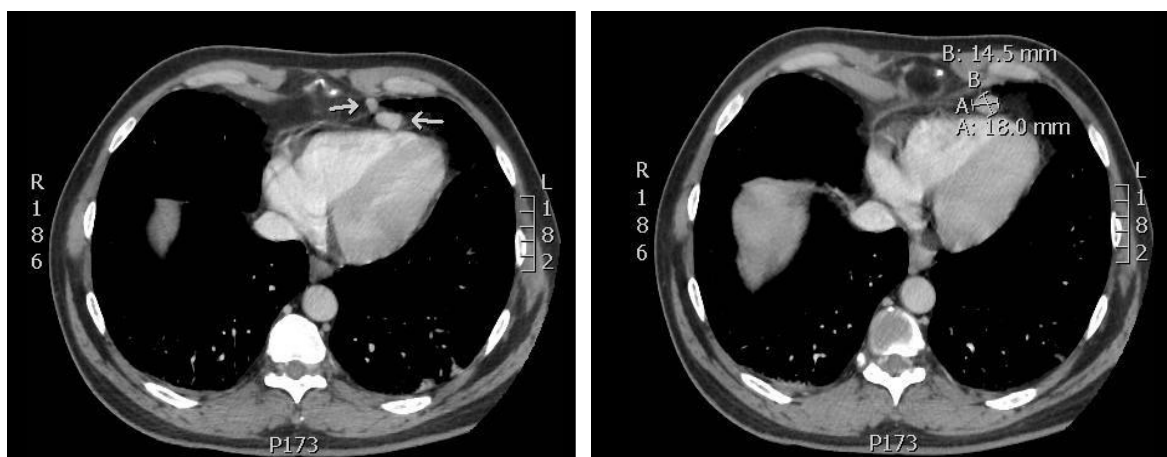
**Discussion**

Buchbinder and Lipkoff were the authors who first describe evidence and mechanism of splenosis in 1939 (4). Splenosis

occurs in up to 67% of patients with a splenic rupture, and it is most commonly localized in the mesentery, peritoneum and omentum (5). Thoracic splenosis is a rare condition and may be found in 18% of patients after a splenic rupture (1,2). However, the real significance is underestimated because most patients are asymptomatic and the diagnosis is incidental. Thoracic splenosis was discov-



**Figure 5a.** CT before administration of intravenous material contrast: nodules in the fat tissue of the parapericardial space.



**Figure 5b (left) – 5c (right).** CT after administration of intravenous material contrast (equilibrium phase): nodules in the the parapericardial space show the same CT features of the nodules localized in the left abdominal cavity.

ered for the first time during an autopsy on a 25-year-old male in Germany in 1896. To our knowledge, only about 40 cases of thoracic splenosis have been reported in the literature (2,3). Splenosis is a different condition from accessory spleens. Splenosis is an acquired condition caused by heterotopic autotransplantation of focal deposits of splenic tissue as a result of trauma or, more rarely, iatrogenic injury involving the spleen; the nodules of splenosis do not have their own hilum or capsule and they receive blood supply from adjacent and surrounding tissues. Accessory spleens are congenital conditions, present in approximately 10% of the population, arising from abnormal spleen embryogenesis, mostly localized in proximity to splenocolic, splenorenal and gastrosplenic ligaments. Accessory spleens have their own hilum and capsule, and they receive blood supply mostly from the small branches of the splenic artery. Most frequently thoracic splenosis follows thoracoabdominal trauma with concomitant lesions of spleen and diaphragm, which allow implantation of splenic tissue most commonly on the left pleural cavity (on parietal or visceral pleura) due to the anatomical position, but other localizations (such as into the lung parenchyma secondary to lung laceration) are described (1,5,6). However, some cases of thoracic splenosis are described in the literature without a diaphragmatic tear but they are attributed to the passage of splenic cells across the pores of the diaphragm to the thoracic cavity (7). In 75% of patients there are multiple nodules, while pleural solitary nodules represent 25% of cases (2), with sizes ranging from few millimeters to 6-7 cm (4). From the time of trauma, the average period for diagnosing thoracic splenosis is approximately 21 years, ranging from 6 to 46 years (2,8). Patients with thoracic splenosis are generally asymptomatic and the condition is discovered incidentally after radiological investigations. However, patients may present symptoms, such as fever, pleuritic pain and haemoptysis. In asymptomatic or symptomatic splenectomized patients who are diagnosed with masses on imaging, phy-

sicians should consider splenosis as differential diagnosis. Other conditions can mimic radiological findings of thoracic splenosis, so physicians must necessarily consider them, such as lymphoma, hamartomas, atelectasis, malignant mesothelioma, invasive thymoma, schwannoma, infectious disease, pleural metastases (most commonly from lung and breast cancer or melanoma), localized fibrosis and scarred pleural lesions. In patients with history of spleen trauma, diaphragm injury and suggestive x-ray or CT images, thoracic splenosis should be suspected and should be investigated also with nuclear medicine studies to confirm the diagnosis without using invasive methods, while in the past when anatomopathological studies were performed obtaining tissue samples by thoracotomy, videothoracoscopy or tru-cut biopsy. Nowadays, imaging investigations allow certain diagnosis (9). In radiological investigations, nodules of splenosis behave similarly to the normal spleen. The CT scan and MR present intense uptake of intravenous contrast material at 70 seconds (equilibrium phase) with a vascular behavior similar to normal splenic tissue. In addition to these investigations, nuclear medicine can confirm thoracic splenosis performing scintigraphy with  $^{99m}\text{Tc}$ -sulfur colloid, scintigraphy with indium-111-marked platelets or scintigraphy with  $^{99m}\text{Tc}$ -labelled heat-damage erythrocytes. The last two methods are preferred due higher sensitivity and specificity. According to the total volume of ectopic splenic tissue, splenic function may recover in part or totally. Therefore, most authors follow a conservative management in cases of splenosis (with clinical and radiological investigations to follow up) because the partial or total extirpation of functioning splenic nodule tissue can increase the risk of new splenosis and infection. In fact, the most serious consequence of splenectomy is increased susceptibility to encapsulated bacterial infections (i.e. by *Streptococcus pneumoniae*, *Neisseria meningitidis*, *Haemophilus influenzae* and some Gram-negative enteric bacteria). Therefore, excision of splenic nodules should be considered if the diagnosis is uncon-

firmed or if patients are symptomatic. However, at least 50% of original splenic tissue is needed for protection against encapsulated microorganisms (10).

### Conclusions

Thoracic splenosis is a benign condition, often incidentally found on chest x-ray, CT scan or thoracoabdominal MR when performed for another reasons. Nodules of splenosis grow slowly after implantation due to trauma with spleen injury and diaphragm tears as well. In splenectomized patients who present unexplained masses on imaging, splenosis should be considered as differential diagnosis. Radiological investigations and nuclear medicine studies can confirm thoracic splenosis without using invasive methods. Excision of splenic nodules should only be considered for symptomatic patients, or if the diagnosis is unconfirmed, because excision increases the risk of new splenosis and infection.

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