# Combined Subcutaneous, Intrathoracic and Abdominal Splenosis

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#### Abstract

We report a case of combined subcutaneous, intrathoracic, and abdominal splenosis who presented with attacks of flushing, tachycardia and vague abdominal pain. The patient's past medical history included a splenectomy due to abdominal trauma and years later, a lung lobectomy due to recurrent pneumonia. An enhancing solid mass adjacent to the upper pole of the left kidney and nodular pleural based lesions in the left hemi-thorax along with nodular lesions in subcutaneous tissue of the left chest wall suggested possible adrenal malignancy with multiple metastases. Histopathologic examination demonstrated benign lesions of ectopic splenic tissue.

Keywords: diagnostic imaging, splenosis

## Introduction

Splenosis usually occurs after a traumatic rupture of the spleen and is defined as auto-transplantation of the splenic tissue to ectopic sites.<sup>1</sup> Most commonly it occurs as intraperitoneal nodules which are found either incidentally or after symptomatic complications, and it may become evident several years after the trauma. Splenosis is mostly an asymptomatic disease that leads to unnecessary investigation in order to differentiate it from other benign or malignant lesions. When multiple sites (with several manifestations) are involved, the situation becomes more complex. The diagnosis is challenging if splenosis is not suspected.

We report a case of combined abdominal, intrathoracic, and subcutaneous splenosis 18 years after a thoracoabdominal penetrating trauma that required splenectomy and diaphragmatic rupture repair.

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### **Case Report**

A 40-year-old man presented with attacks of flushing, tachycardia, and vague abdominal pain. His physical examination was unremarkable and his blood pressure was normal. Hyperglycemia was obvious in his biochemical exam. There was no drug abuse history. The patient's past medical history was remarkable for two previous surgeries; a splenectomy following a gunshot wound which passed the left abdominal upper quadrant into the thorax 18 years ago and 13 years later, a left lung lower lobe lobectomy because of recurrent pneumonia due to a foreign body (bullet) and repair of diaphragmatic tearing which was mismanaged at the time of the trauma.

Further evaluation with contrast-enhancing abdominal CT scan revealed an enhancing solid mass  $(70 \times 40 \times 55 \text{ mm})$  adjacent to the upper pole of the left kidney (inseparable from the adrenal) and its mass effect on the left kidney (Figure 1). The CT scan also identified some similar nodular pleural based lesions in the left hemi-thorax just above the repaired hemi-diaphragm as well as nodular lesions

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Figure 1. CT scan of the abdomen showing an enhancing solid mass adjacent to the upper pole of the left kidney and its mass effect



Figure 2. Some nodular pleural based lesions in the left hemi-thorax just above the repaired hemi-diaphragm



Figure 3. Nodular lesions in subcutaneous tissue of the left chest wall



Figure 4. Histopathologic view of abdominal mass (H&E 40x).



Figure 5. The nodular pleural based lesions in the left hemi-thorax just above the left hemi-diaphragm

in subcutaneous tissue of the left chest wall which suggested a possible adrenal mass with multiple metastases to the pleura and left hemi-diaphragm (Figures 2 and 3).

Subsequently, the patient underwent a surgical procedure to exclude adrenal neoplasm (most likely pheochromocytoma). No defect was observed in the left hemi-diaphragm, it was thin and eventrated.

Histopathologic examination confirmed splenic tissue (Figure 4). A needle biopsy from the thoracic nodules and excisional biopsy from the subcutaneous nodule confirmed ectopic splenic tissue in both. A second look at the chest CT scan at the time of the left lower lobectomy detected similar pleural based nodules, misinterpreted as pleural thickening and previous reaction (Figure 5).

### Discussion

Splenosis is a rare condition.<sup>2</sup> Peritoneal splenosis is estimated to occur in up to 67% of patients with splenic rupture followed by splenectomy<sup>3</sup> and in 18% following diaphragmatic rupture.<sup>4</sup> The time interval between the injury and the diagnosis ranges from 1 to 42 years, with an average of 18.8 years.<sup>5</sup> The basic pathogenesis is believed to be a mechanical implantation of splenic tissue, most commonly on peritoneal or serosal surfaces, following a traumatic or surgical injury of the splenic capsule.<sup>3</sup> The splenic pulp is able to implant itself onto the serosal surfaces of the chest, abdomen or pelvis to derive its own blood supply from the submesothelial vasculature or from the surrounding circulation (for example, pulmonary circulation) and grow into histologically mature splenic tissue.<sup>4</sup>

Intrathoracic splenosis is rare and an accompanying diaphragmatic defect is an essential part of its pathogenesis. It is usually asymptomatic (rarely pleurisy or recurrent hemoptysis), in contrast to abdominal splenosis in which abdominal pain, intestinal obstruction, gastrointestinal bleeding and the recurrence of hematologic disorders have been described.<sup>5</sup>

Chest CT reveals a solitary pleural-based nodule or mass in 25% of cases and multiple pleural-based nodules in the remaining 75%.<sup>2</sup> Pleural-based implants may occur on the parietal or visceral pleura, and they have similar attenuation to that of the normal spleen.<sup>2</sup> The present patient had multiple pleural-based nodules, vague abdominal pain and flushing due to suprarenal splenosis. Differential diagnoses included pleural metastases (probably arising from lungs, breasts, or a melanoma), lymphoma and localized fibrous tumor of the pleura, malignant mesothelioma or invasive thymoma.

Subcutaneous splenosis is extremely rare. To the best of our knowledge, only 11 cases have been reported in the literature with histories of splenic rupture. Auto-implants of splenic tissue were found in old abdominal surgical scars in eight cases, whereas three cases presented at the site of an exit gunshot wound scar. In our patient, subcutaneous splenosis was also found in a scar at the site of an exit gunshot wound.<sup>3</sup>

Splenosis of multiple sites results in a complex situation<sup>6,7</sup> which requires intensive clinical and radiological investigations to avoid unnecessary surgeries. Previous thoracoabdominal trauma and ruptured spleen pointed to a diagnosis of splenosis in our patient. A scintigraphic examination could be performed not only to avoid unnecessary surgery but also to preclude removal of the ectopic spleen.

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