

Thoracic Splenosis More Than 40 Years After Thoracoabdominal Trauma

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Financial Disclosures:
None reported.

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Submitted
February 4, 2013;
final revision received
May 14, 2013;
accepted
May 31, 2013.

Splenosis is a rare occurrence that is defined as autotransplantation of splenic tissue usually after splenic rupture due to trauma and subsequent splenectomy. Although splenosis most commonly occurs in the abdomen, the authors report a rare case of thoracic splenosis after remote thoracoabdominal trauma. A 62-year-old woman was found to have lower-lobe, pleural-based nodular lesions in juxtaposition to the posteromedial segment of the lung during workup for an abdominal hernia. Surgical excision of the mass confirmed the diagnosis of ectopic splenic tissue, and splenosis was diagnosed. This woman was among the rare 18% of people who are found to have splenosis in the intrathoracic space. In the workup of pulmonary nodules in patients with a history of trauma, splenosis should be a consideration.

J Am Osteopath Assoc. 2013;113(11):853-856
doi:10.7556/jaoa.2013.061

Splenosis, a condition also known as ectopic spleen, is an extremely rare occurrence that is defined as autotransplantation of splenic tissue usually after splenic rupture due to trauma and in association with a subsequent splenectomy.¹ With splenosis, splenic tissue is most commonly seeded into the abdominal cavity or pelvis, while thoracic implants make up only 18% of the cases of splenosis.¹ Thoracic presentation is most commonly an incidental finding, and the patient is usually asymptomatic.² Patients may also present with a palpable abdominal mass.³ Computed tomographic (CT) or ultrasonographic imaging should be used to identify areas of possible ectopic tissue, although diagnosis is confirmed postoperatively by means of pathologic analysis. We report a case of thoracic splenosis found more than 40 years after a patient's initial trauma.

Report of Case

A 62-year-old morbidly obese woman with a medical history of atrial fibrillation presented to the university general surgery division for workup of an abdominal incisional hernia. A CT scan of the hernia was obtained, and a pleural-based mass was noted. The patient was referred to the cardiothoracic division for evaluation of the mass. The patient denied dyspnea, history of weight loss, or previous workup for hernia. Surgical history was notable for cholecystectomy, gastric bypass surgery, and emergency splenectomy with

repair of the lower lobe of the left lung with chest tube placement at age 19 years secondary to a close-range gunshot wound.

A CT scan of the left side of the chest was obtained, and it revealed a lower-lobe, pleural-based nodular lesion in the posteromedial and posterior part of the left lung (*Figure 1*).

Although the patient's history included neither neoplasm nor tobacco use, the lesions were suggestive of metastases, and the radiologist recommended positron emission tomography. The scan revealed no other suspicious lesions and was not indicative of hypermetabolic activity in the thorax. Other differential diagnoses included splenosis, mesothelioma, and residual scar tissue as a result of traumatic lung repair. Osteopathic structural examination revealed tissue tenderness throughout the left costophrenic region and restriction of the left lower ribs.

A video-assisted thoracoscopy of her left side revealed a fleshy mass of 2 to 3 cm along the left dia-

phragmatic surface. Resection of the mass—with repair of the resultant diaphragmatic defect—and wedge resection of the representative section of scarred lower lobe of the left lung were performed. We noted clinically significant intrathoracic inflammatory changes within the thoracic space, which were consistent with the patient's history of previous chest intubation. Pathologic results revealed the fleshy red mass to be consistent with benign splenosis (*Figure 2*). A 22F left-sided pleural chest tube was left in place, and the patient was transferred to the intensive care unit with ventilatory support. She was extubated and transferred to the step-down floor on postoperative day 1. The chest drain was removed on day 2, and the patient was discharged the following morning. Osteopathic manipulative treatment consisted of balanced ligamentous tension, indirect myofascial release of both the sternum and the respiratory diaphragm, and Sibson fascial release, as well as rib raising, with attention to the cervical and thoracic spine, as described by O-Yurvati

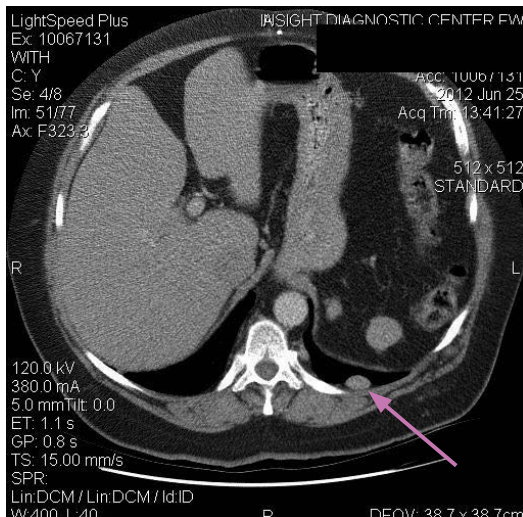


Figure 1. Axial view from a computed tomographic image revealing a pleural-based mass (arrow) in a 62-year-old woman. The patient underwent emergency splenectomy with repair of the left lower lobe of the lung and with chest intubation at age 19 years secondary to a close-range gunshot wound.

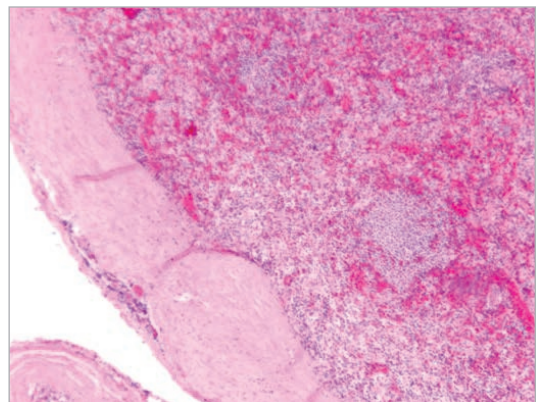


Figure 2. Hematoxylin and eosin stain of splenic tissue from a biopsy in a 62-year-old patient who had undergone emergency splenectomy at age 19 years.

et al.⁴ The patient's condition improved postoperatively, and she was followed up in the office 2 weeks after surgery without any apparent complications.

Comment

Splenuis is most commonly attributable to penetrating or blunt-force trauma to the thoracoabdominal region. As imaging technology has improved, splenuis has been noted in up to two-thirds of patients after splenuectomy for trauma.⁵ Furthermore, after splenic rupture, splenuis is relatively common, with a rate of up to 65% according to a case study by Malik et al.⁶ The majority of documented cases of splenuis after trauma have been noted in the intrabdominal space, most commonly in the peritoneum, omentum, and mesentery. For the 18% of patients who have splenuis localized to the thorax, the most common ectopic site is the pleural space of the left lung.^{6,7}

In cases of splenuis, splenic tissue can be transferred into the thoracic cavity by either hematogenous spread or by passing through a tear in the diaphragm.⁷ The etiologic process of splenuis includes splenic trauma or splenuectomy⁸; splenuis occurs less commonly as a result of congenital malformations from incomplete fusion of the dorsal mesogastrium.³

Imaging of thoracic splenuis can be made noninvasively by means of scintigraphy with technetium Tc 99m heat-damaged erythrocytes.⁶ Noninvasive heat-damaged scintigraphy has the highest specificity of all nuclear imaging diagnostic modalities, and some observers regard it as the standard for splenuis.⁹ If scintigraphy is unavailable or results are inconclusive, further diagnosis can be achieved with fine-needle aspiration. Unfortunately, the location of a lesion often precludes fine-needle aspiration; if a lesion is accessible, its pathologic presentation can be misleading, with lymphocytic infiltrate misdiagnosed as lymphoma.⁶ Video-assisted thoracoscopic surgery is an option that can serve both diagnostic and therapeutic purposes. Pathologic analysis

can rule out such causes as pulmonary metastases, non-Hodgkin lymphoma, or mesothelioma.

If the diagnosis can be confirmed preoperatively, surgery is not indicated unless the patient is symptomatic.⁹ It is usually not necessary to remove the pulmonary nodules because the splenic tissue is slow growing, noninvasive, and nonmalignant. The splenic tissue that is found in the lungs resembles tissue that is found in the spleen—with areas of red pulp, white pulp, and lymphoid follicles—and is usually surrounded by a capsule and fibrosis resulting from thoracoabdominal trauma. As was shown in the present case, most cases of thoracic splenuis are found incidentally by means of CT or magnetic resonance imaging because the majority of patients are asymptomatic. To our knowledge to date, there have been 4 reported symptomatic cases of thoracic splenuis; 2 patients reported having hemoptysis, 1 patient complained of a productive cough, and 1 patient reported having pleuritic chest pain.⁶

The most serious complication in patients who have undergone splenuectomy are increased infection rates and sepsis with encapsulated organisms such as *Neisseria meningitidis*, *Streptococcus pneumoniae*, and *Haemophilus influenzae*. Some physicians have postulated that maintaining even a small amount of splenic tissue after splenuectomy may offer immunologic advantages.¹⁰ In 2006, Backhus and Bremner¹⁰ suggested normal immune function following splenuectomy due to splenuis tissue leading to a lack of Howell-Jolly bodies, "pitted" erythrocytes, and siderocytes in the peripheral blood smear.¹¹ However, since 2006 many animal studies and human anecdotal data have shown that splenic nodules, when separated from the spleen itself, do not regain full immunologic capability.¹² Patients with sepsis and splenuis have an estimated 58 times increased mortality or higher as compared with individuals with normal spleen function.¹² Such differences may be attributed to splenic implants resulting from cellular growth rather than redistributed portions of disrupted spleen. Furthermore, ectopic splenic tissue has a decreased amount of white

pulp.¹² The functionality of ectopic splenic tissue continues to be strongly debated.

Postoperative OMT as described by O-Yurvati et al⁴ can be performed safely in patients with splenosis, and, with OMT, normal recovery may be expected.

Conclusion

Although thoracic splenosis is a rare occurrence, patients who present with 2 factors—several asymptomatic left pleural-based pulmonary nodules and a history of thoracoabdominal trauma with splenectomy—should be considered for such a diagnosis. Interestingly, the average interval between initial trauma and diagnosis of thoracic splenosis is more than 18 years.⁹ Our patient presented more than 40 years after her initial traumatic event. Clinical history is invaluable in patients with similar presentations. In workup, scintigraphy is a valuable tool, if available, and the images that it yields may preclude thoracotomy.¹³ A minimally invasive approach for excision of such masses can be completed with minimal morbidity. In this case, it is likely that the patient developed splenosis secondary to seeding by her previously placed chest tube after splenic trauma. In the workup of pulmonary nodules or masses in patients with a history of trauma, splenosis should be included in the differential diagnosis.

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