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Case Report

A case of thoracoabdominal splenosis

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ABSTRACT

We describe a case of a 38-year-old male with a remote history of motor vehicle trauma who presented to the emergency department with 1-week history of progressively worsening abdominal pain localized to the epigastric region. Patient history included splenectomy. Computerized tomography demonstrated multiple masses in the left pleural space as well as masses continuous with the diaphragm and abdominal wall in the left upper quadrant. In addition, a lobulated mass was identified in the right upper quadrant along the anterior right hepatic lobe. A diaphragmatic defect was noted containing splenic tissue. A diagnosis of splenosis was made. Disseminated splenosis presenting in both the thorax and abdomen is rare and poorly documented. This case serves to further illuminate this condition.

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Introduction

Splenosis is characterized by the ectopic transplantation of splenic tissue, often as a result of trauma [1]. Splenosis has a different pathophysiology than accessory spleens, which result not from a traumatic etiology but from incomplete fusion of mesenchymal buds during embryogenesis [2]. Accessory spleens are estimated to occur in 10%-30% of the general population [2], while splenosis may be found in about a quarter of patients undergoing splenectomy for trauma [3]. Since splenosis often presents asymptotically, it is typically an incidental finding on imaging [4]. The mean time between the

injury responsible for splenosis and the actual diagnosis is about 2 decades [5]. Nevertheless, there are reports of splenosis presenting as persistent chest pain [6,7], hemoptysis [8], and bowel obstruction [9]. Although diagnosis via biopsy is the gold standard, imaging informed by a relevant patient history including splenectomy or prior trauma to the spleen, is still acceptable [10].

Splenosis is most prevalent in the abdomen and, much less commonly, in the thorax [6]; here, we report a case of disseminated asymptomatic splenosis localized to the left pleural cavity and upper right and left quadrants in a 38-year-old male with a remote history of motor vehicle trauma.

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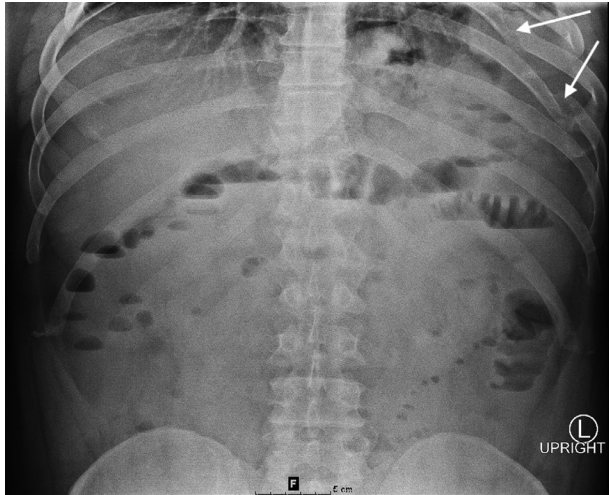


Fig. 1 – Radiograph of abdomen, upright anteroposterior projection, of a 38-year-old male, demonstrating chronic rib fractures (arrows) on the left with nonspecific bowel gas pattern.

Case report

A 38-year-old male with a history of motor vehicle trauma presented to the emergency department with abdominal pain that had been ongoing for 1 week. The patient reported the pain was localized to the epigastric region, had been progressively worsening, and was not modifiable. That morning, the patient vomited once after eating and noted that his abdominal pain was especially severe. He admitted to having dark but not tarry stools. He denied fever and had no recent travel history. The prior day, the patient had been seen at another ER, where he was diagnosed with acid reflux and prescribed omeprazole.

The patient's past medical history was notable for splenectomy and 2 skin grafts on his left arm owing to a remote history of motor vehicle trauma. In addition, the patient also had an abdominal wall hernia repair. The patient is an every day smoker and consumes alcohol on a weekly basis.

On physical exam the patient appeared mildly in distress. Pulse was 103, BP 149/88, and temperature 97.9 F. Abdomen was nondistended, soft, but marginally tender to palpation in the epigastric region. The patient was not in respiratory distress; lungs were clear to auscultation bilaterally.

Given the abdominal pain and vomiting, an initial 2-view abdominal x-ray series was conducted and showed multiple air fluid levels in the small bowel (Fig. 1). A follow up computerized tomography (CT) of the abdomen/pelvis with contrast indicated several left-sided chronic rib fractures, nonspecific bowel gas patterns; fluid present in the colon; a prominent appendix, otherwise normal in appearance, with an appendicolith. Small bowel obstruction was suspected. Incidentally, the CT also revealed multiple lobulated masses, identified as splenules, in the left pleural space, the largest of which mea-

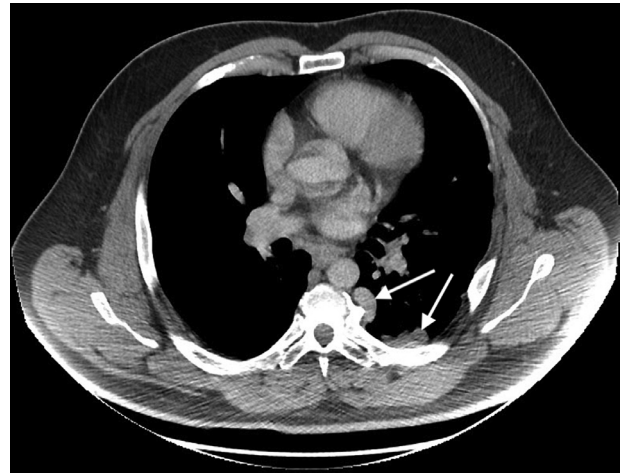


Fig. 2 – CT enhanced image, axial projection, displaying nodularity (arrows) of left pleural space. CT, computed tomography.

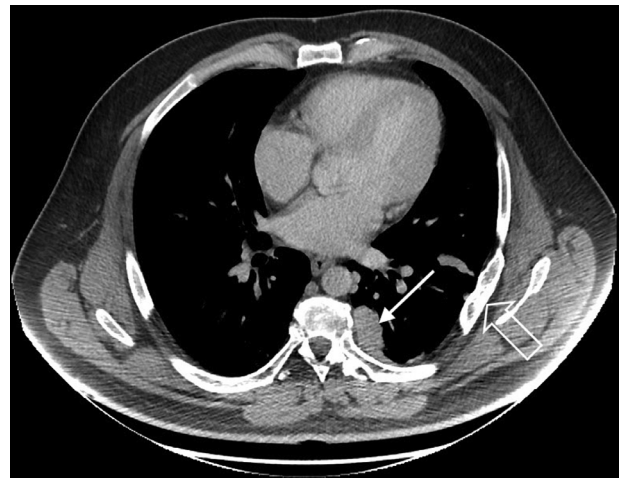


Fig. 3 – CT enhanced image, axial projection, displaying nodularity (solid arrow) of left pleura space and chronic deformity of an adjacent rib (open arrow) from prior trauma.

sured about 4 cm in diameter (Figs. 2 and 3). Splenules, which measured at most 3.3 cm in diameter, were observed continuous with the diaphragm and abdominal wall in the left upper quadrant and were noted traversing a diaphragmatic defect (Figs. 4–6). In addition, a splenule was identified in the right upper quadrant along the anterior right hepatic lobe, no less than 2 cm in diameter.

The patient was provided IV saline (1 L) and ondansetron for his nausea and vomiting. The imaging results were shared with the patient. Discharge instructions included continuation of omeprazole and ondansetron with a referral for additional work up to rule out pleural malignancy.

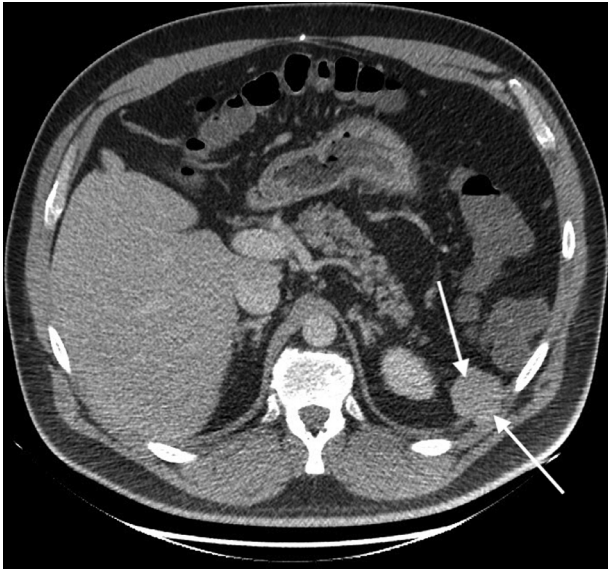


Fig. 4 – CT enhanced image, axial projection, displaying splenosis (arrows) traversing a diaphragmatic defect.



Fig. 5 – CT enhanced image, axial project, displaying splenosis (arrows) on both sides of the diaphragm.

Discussion

The patient presented in this report is a 38-year-old male who was diagnosed via imaging with disseminated splenosis localized to the left pleural cavity and upper left and right abdominal quadrants. To our best knowledge, disseminated splenosis in both the thorax and abdomen is rare and has only been published once in the English language literature. Sanchez et al at the Hospital Universitario Virgen del Rocio in Spain documented a case of thoracoabdominal splenosis in a 54-year-old male with a history of chest trauma [11]. The largest splenosis

found in our patient measured up to 3-4 cm in diameter, which is consistent with the literature [12,13].

Splenosis may masquerade as malignancies, including carcinomatosis [14], liver tumor [15], metastatic right adrenal mass [16], lymphoma [17], and lung cancer [18]. When compounded by risk factors like smoking, an accurate diagnosis of a pleural mass as thoracic splenosis can be difficult [19]. Important clues in our case were – patient history of prior trauma and splenectomy, and imaging results showing left-sided chronic rib fractures with diaphragmatic transgression and masses located in the left hemithorax. Damage to the diaphragm may be essential for splenosis to present

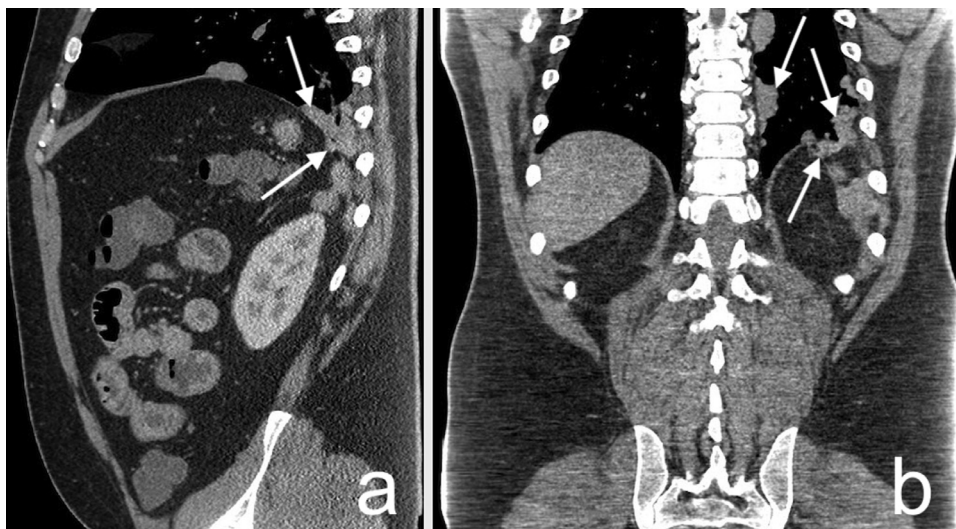


Fig. 6 – CT enhanced images. (a) Sagittal and (b) coronal projections demonstrating splenosis (arrows) on both sides of the diaphragm.

in the pleural space [20]. Moreover, the age of the patient must also be considered. While the 38-year-old male is a daily smoker, the median age of lung cancer diagnosis is 70 years old [21], making pleural malignancy a less likely diagnosis. Although we did not conduct nuclear imaging, Technetium-99m heat damaged erythrocyte study is considered the optimal imaging technique for splenosis [22], we are confident in our diagnosis because the pleural masses and the left upper quadrant masses are continuous across the diaphragmatic defect.

Patients presenting with splenosis, by definition, have experienced trauma to the spleen and or splenectomy. Since the primary purpose of the spleen is to filter aging erythrocytes and blood borne pathogens [23], asplenic patients are at increased risk for infections, particularly from *Hemophilus influenza*, *Streptococcus pneumonia*, and *Neisseria meningitis*—all encapsulated bacteria that are typically eliminated by a functioning spleen [24]. Asplenic patients are also at heightened risk for developing sepsis [25]. While some reports indicate splenosis may provide immune function [26,27], a review of the literature by Connell et al suggests that such benefit, if it exists, is not protective against overwhelming infection like sepsis [10]. Our patient did not present with any history of infections or sepsis.

In terms of treatment, asymptomatic splenosis does not warrant surgical intervention [20]. However, we were unable to find any prospective studies evaluating the long-term outcomes of patients diagnosed with splenosis. Further investigation is necessary to better understand the sequela of splenosis.

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