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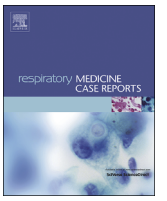

Case Report

Posttraumatic thoracic splenosis in a 77-year-old patient after a motor vehicle accident

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ABSTRACT

We present a rare case of autotransplantation of splenic tissue to the thoracic cavity, concerning for lung tumor, after splenic and diaphragmatic injury following a severe motor vehicle accident 27 years previous.

1. Introduction

Thoracic splenosis, involving autotransplantation of splenic tissue to the thoracic cavity after splenic and diaphragmatic injury, is rarer than abdominal splenosis [1,2]. We describe a case of posttraumatic thoracic splenosis, concerning for lung tumor, in a patient decades after severe motor vehicle accident, and review extant literature reported cases.

2. Case presentation

A 77-year-old male with a history of severe thoracoabdominal trauma presented with exertional dyspnea and nonpleuritic chest wall pain. Severe motor vehicle accident 27 years previous was complicated by pneumothoraces, requiring multiple chest tubes. The patient was a non-smoker with a history of agent orange exposure from military service in Vietnam.

Initial evaluation revealed normal myocardial perfusion stress test. Chest computed tomography (CT) showed multiple pleural base nodules along the left posterior and posterolateral chest wall, measuring up to 34 mm (Fig. 1a–e), chronic left rib fractures, and a small area of splenic tissue beside a large cystic mass in the left upper quadrant (Fig. 2). Of note, areas of lobulation were near previous chest tube placement and rib fractures with malunion. Breathing treatments were initiated. PET scan demonstrated no abnormal uptake, with maximum SUV of 2.3 in the lobulated areas of the left hemithorax. Given PET scans with upper limit of normal uptake and his previous exposure history, he underwent a CT-guided percutaneous needle biopsy along the pleura. Microscopic examination of the pleural lobulation biopsy specimen confirmed splenic tissue (Fig. 3). The combination of clinical data, imaging, and biopsy allowed the diagnosis of posttraumatic thoracic splenosis with no surgical intervention. He maintained outpatient medical management with active surveillance.

Abbreviations: CT, computed tomography; PET, positron emission tomography; SUV, standardized uptake value; SPECT, single photon emission computed tomography; VATS, video-assisted thoracoscopic surgery; ITP, immune thrombocytopenic purpura; MRI, magnetic resonance imaging; MVA, motor vehicle accident; GSW, gunshot wound; Tc, technetium.

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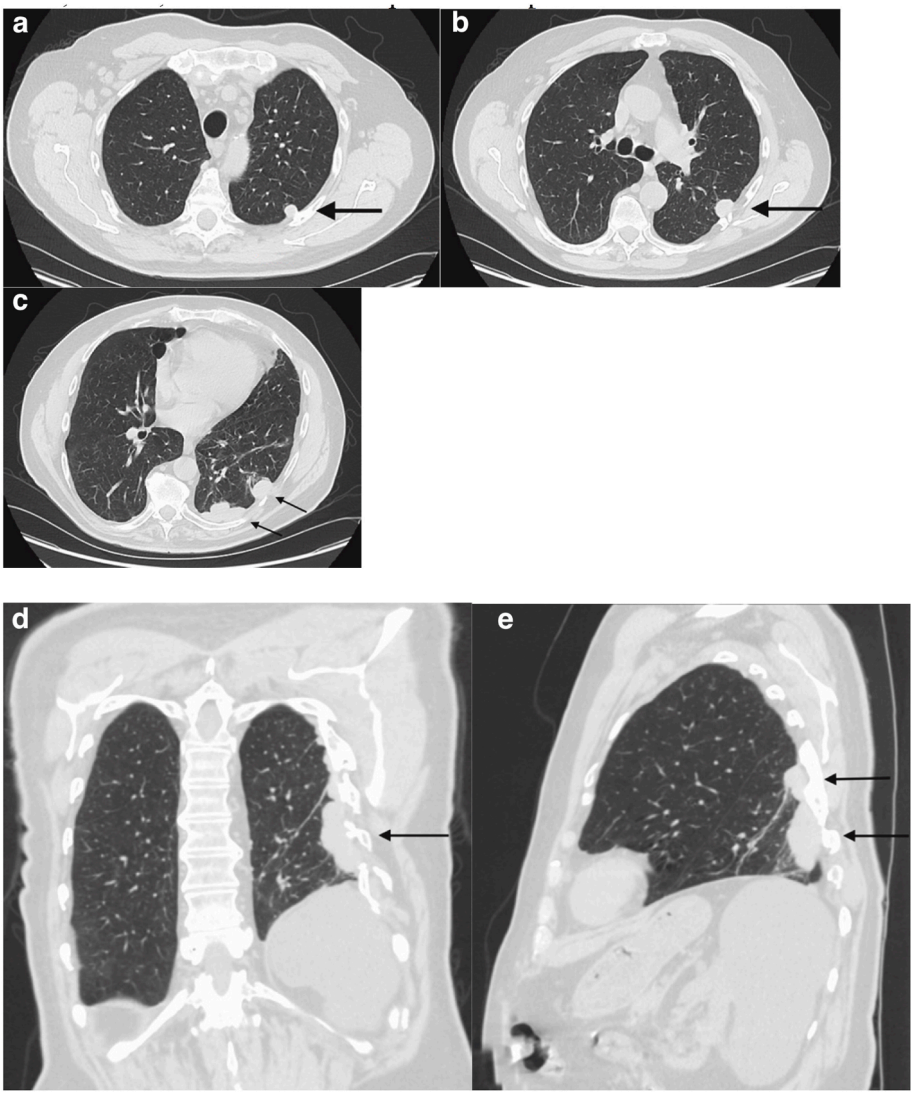

Fig. 1. CT axial (a, b, c), coronal (d), and sagittal (e) images showing numerous pleural base nodules along the left posterior and posterolateral chest wall (*arrows)*. Sizes ranged from 1.2 cm, 2 cm, 3.4 cm, 5 × 2.4 cm on the posterior aspect of the LLL.

3. Discussion

Thoracic splenosis is a rare phenomenon where splenic tissue autotransplants into the thoracic cavity. The interval between trauma and time of clinical presentation can range from less than a year to 42 years [3]. One mechanism contributing dissemination is splenic pulp seeding the adjacent cavity [4]. Less common mechanisms, such as hematogenous spread of splenic pulp and hypoxia induced tissue growth, have been suggested as well [4]. Migration of splenic tissue into the chest cavity occurs less frequent, 18% [5], compared to intrabdominal implantation, almost 67% of reported splenosis cases [6,7]. Almost all reported cases of thoracic splenosis are associated traumatic injury to the abdomen, as in the current case, requiring splenectomy with or without hemi-diaphragmatic repair as demonstrated in Table 1.

In general, splenic tissue implants onto the serosal surface of the thoracic cavity after seeding through an opening in the diaphragm and progresses to nodules [8]. Nodules then derive their blood supply from the surrounding tissue and vessels [9]. Predictably, implantations which typically occur in the left hemithorax after injury to the diaphragm due to trauma or after intrabdominal procedures have been associated with the anatomical position of the spleen [3]. Serosal implants are more commonly seen than parenchymal implants, which are far less common.

Unlike abdominal splenosis which typically is asymptomatic, thoracic splenosis is often diagnosed when patients present with nonspecific pulmonary, esophageal or cardiac symptoms [10,11]. Symptoms such as hemoptysis and pleurisy are uncommon [10].

Diagnosis often requires imaging modalities which are able identify reticulo-endothelial specific uptake, which helps differentiate thoracic splenosis from malignancies [12]. The most sensitive and specific modality is 99 m technetium-labeled colloid scan paired

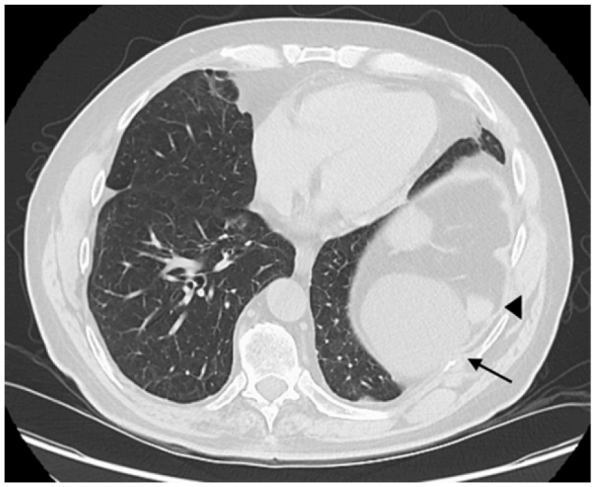

Fig. 2. Small area of splenic tissue (*arrowhead)* beside a large cystic area in the LUQ in place of the spleen, 13.7 cm (*arrow*).

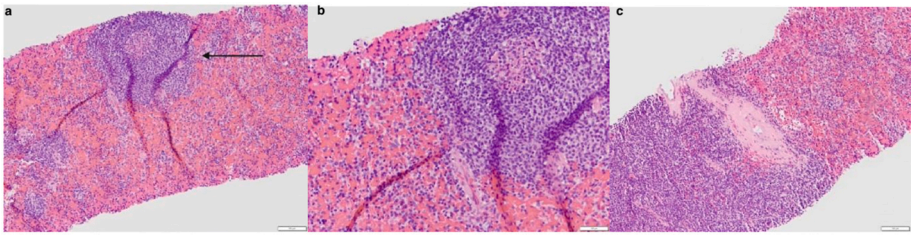

Fig. 3. Microscopic examination of the pleural lobulation confirmed splenic tissue, including white pulp (*arrow*) embedded in a red matrix (red pulp). Scale: (a,c) = 4× magnification, (b) = 20× magnification. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

with single photon emission computed tomography/computed tomography (SPECT/CT) [13]. The role of PET scan in identifying splenic tissue is limited, as it is not specific.

Table 1 describes 12 cases of reported splenosis in the past 10 years including the current case. Among the 11 cases, 58% were male, with a sex ratio of 1:0.71. Average age in the series was 53 (range 38–81). The most common mechanisms of injury were MVA, GSW and blunt abdominal trauma. As demonstrated, most patients present with multiple nodules with max sizes varying from 18 to 100 mm predominantly in the left hemithorax which could be due to the anatomy. Average time from injury/splenectomy to diagnosis was 27 years. Literature review also shows that thoracic splenosis almost always occurs after injury to the left abdomen leading to splenic rapture. Placement of chest tube as the primary suspected cause of thoracic splenosis is unique to our patient. In most cases, the predominant symptoms included cough and chest pain with few cases presenting with fever, weight loss and dysphagia [10]. Often, splenic tissue did not require removal, but there is limited data which suggests that removal may be beneficial in patients with persistent Immune Thrombocytopenic Purpura (ITP) [22]. Although 2 out of the reported case series required removal of at least one mass, no additional surgical intervention was required or performed after pathological diagnosis. Only one patient was found to benefit from removal of mass as it was compressing the esophagus [10,16].

Additionally, video-assisted thoracoscopic surgery (VATS), as well as imaging guided biopsy, is often unnecessary in most cases [14]. Technetium-99m (Tc-99m) heat-damaged erythrocyte nuclear imaging, together with detailed history, is the gold standard for diagnosis [4]. Alternatives to this modality include Tc-99m sulfur colloid and indium-111-labeled platelets and ferumoxide Magnetic Resonance Imaging (MRI) [15].

Although true in most instances, diagnosis becomes challenging when patients have a history of cancer, which may warrant additional testing, such as tumor markers [12,16]. As demonstrated in Table 1, splenic tissue is often found tethered to the diaphragm or closes to well vascularized organs in the thoracic cavity, such as the pericardium, thoracic wall, and diaphragm. Although the exact mechanism of revascularization has not been fully explored, its embryonic formation offers some clues [17].

Our patient presented with symptoms of exertional dyspnea and nonpleuritic chest pain. Detailed review of history revealed thoracoabdominal trauma after a MVA 27 years prior to presentation. Although the patient underwent CT-guided biopsy, we strongly believe this could have been avoided with detailed history and 99m-Tc with PECT/CT imaging. Unique to this patient is the presence of thoracic splenosis after chest tube placement, unlike most patients who present after abdominal trauma requiring splenectomy. Like most patients diagnosed with thoracic splenosis, our patient did not require any treatment. Dyspnea was unrelated to the mass found during admission.

Table 1
Review of the clinical characteristics of 12 patients with thoracic splenosis.

References	Gender/ age	Risk factor	Time of injury to Dx (yrs)	Max Size (mm)	Location	Number of nodules	Initial suspected lesion	Symptoms	Diagnostic Modality
[3] Lopes 2014	F/54	Bomb explosion, s/p splenectomy	40	62	L. mediastinal and juxtapleural nodules	Multiple	Bacterial PNA	Cough, yellow sputum, dyspnea, wheezing, Fever 39 °C	99m-technetium (99m-Tc) stain colloid scintigraphy
[10] Ha 2019	M/53	MVA, S/p splenectomy via laparoscopic	15	50	L.EG junction	Single	Lymphoid hamartoma, GIST	Chronic heartburn, cough	VATS w/ complete resection of mass from R side
[11] Sonmez 2022	F/39	GSW to abdomen, S/p splenectomy	19	38	L posterior costal pleura and diaphragmatic pleura	Multiple	Pleural tumor	Severe L. chest pain	VATS w/biopsy, 99m-technetium (99m-Tc) stain colloid scintigraphy, MRI
[12] Gelsomino 2016	F/51	Thoraco-Abdominal trauma, S/p splenectomy + nephrectomy	30	45	Paracardial mass, L Costophrenic angle nodule	Multiple, 2	Metastatic breast Ca, S/p radical left mastectomy	Evaluation of adjuvant therapy, Asymptomatic	Tc-99-labeled nanocolloid scan w/fused SPECT-CT
[13] Baldolli 2017	M/50	MVA, S/p splenectomy	34	30	L. Supra- and Subdiaphragmatic lesions	Multiple	Pleuropneumonia	Fever, Chills: (+) CMV	Tc-99m colloid scintigraphy SPECT: Blood smear: (+) Howell-Jolly bodies
[14] Soraru 2015	M/81	Blunt abdominal trauma, s/p splenectomy, nephrectomy, hemi diaphragmatic repair	30	32	Anterior mediastinal, left posterior subpleural nodule	Multiple, 2	Neoplasm	Weight loss, fever, elevated liver enzymes	Tc-99m colloid scintigraphy SPECT, w/o biopsy
[16] Niu 2018	M/44	Fall, s/p splenectomy	10	18	L. Lung, diaphragm, mediastinal pleura	Multiple	Lung Ca	Thoracalgia	VATS w/removal of masses
[18] Remtulla 2017	M/38	MVA, s/p splenectomy via laparotomy	20	41	L. lower lung lobe, posterior, L. ventricle	Multiple	Lung Ca	Dry cough	VATS w/biopsy
[19] Le Bars 2020	M/39	Traumatic splenic rupture	36	100	L. lower pleural space	Single Large, multiple small nodules	Lung Ca	Chronic chest pain	L. Thoracotomy
[20] Thampy 2018	F/48	GSW to abdomen, S/p splenectomy	20	26	L. pleural hemithorax	Multiple	Breast Ca metastasis	L sided shooting CP	99m-technetium (99m-Tc) stain colloid scintigraphy
[21] Williams 2017	F/62	MVA, S/p splenectomy	43	46	Retrocardiac, L. pericardium, L upper quadrant	Multiple	Malignancy	Severe L CP and back pain	99 m technetium-labelled sulphur colloid scan with single photon emission computed tomography/ computed tomography (SPECT/CT), blood smear: (+) Howell-Jolly bodies
Current case	M/77	MVA, s/p multiple chest tube placement w/o splenectomy	27	50	L. posterior and posterolateral hemithorax	Multiple	Pleural tumor	Exertional dyspnea and nonpleuritic chest wall pain	CT chest, PET scan, CT-guided percutaneous needle biopsy

In conclusion, thoracic splenosis has a low incidence among the general population. It often occurs after traumatic abdominal injury with or without obvious diaphragmatic rupture. Patients often present with symptoms which can raise suspicion for pneumonia, esophageal disorders, tumors of the thoracic space, and metastatic lesions. The majority of reported cases do not require treatment, except in cases when nodules cause persistent symptoms [10]. Although VATS has been relied on in some cases to diagnose or rule out malignancies, it is often unnecessary and can lead to further complications. Detailed history of trauma and splenectomy, in combination with radionuclide imaging, is sensitive and specific in diagnosing thoracic splenosis and prevents the burden of surgical procedures. This case provides additional resources in the awareness campaign for accurate thoracic splenosis diagnosis which can facilitate avoidance of unnecessary surgical interventions. We present this case report and case studies in the hope that thoracic splenosis would be part of the differential in patients who present with left hemithoracic mass with a history of abdominal trauma requiring splenectomy.

Declaration of competing interest

Author(s) have participated in this manuscript and have no conflict of interest to report. This paper is not under consideration in any other journal. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the consent is available for review by the Editor of this journal on request. Author(s) agree to transfer manuscript copyright to the journal in exchange for publication. Author(s) defer to the journal for reviewers.

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