

Symptomatic isolated thoracic splenosis 11 years after abdominal trauma – Case report

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SUMMARY

Introduction Thoracic splenosis is defined as the autotransplantation of splenic tissue into thorax. It occurs due to splenic rupture in association with a diaphragmatic tear on the left side after a traumatic event. It is a rare disease that most commonly remains undiscovered as it is usually asymptomatic.

Case Outline We present a symptomatic case of thoracic splenosis in a 53-year-old smoker male patient with a medical history of abdominal surgery and splenectomy for a thoracoabdominal gunshot. Three years before the medical examination he was suffering from dyspnea, frequent coughing, left pleuritic chest pain and complained about faster fatigue. A chest radiograph obtained during a medical checkup showed a multinodular left pleura-based mass in the upper lobe. Established histopathological diagnosis after surgical removal of the nodule was splenosis. No evidence of malignancy was observed.

Conclusion Splenosis should be considered as a differential diagnosis by the undertaken workup of left pulmonary nodules or masses in patients with a history of trauma.

Keywords: symptomatic thoracic splenosis; splenectomy; thoracoabdominal gunshot

INTRODUCTION

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. Thoracic splenosis occurs less frequently than abdominal splenosis and may be found in 18% of patients after splenic rupture. The diagnosis of thoracic splenosis can be established noninvasively with several diagnostic modalities [2, 3]. Computed tomography (CT) or ultrasonographic imaging should be used to identify areas of possible ectopic tissue, although diagnosis is confirmed postoperatively by means of pathologic analysis. A minimally invasive approach for excision of such masses could be completed with minimal morbidity. Video-assisted thoracoscopic surgery is an option that can serve both diagnostic and therapeutic purposes [4]. Pathologic analysis can rule out such causes as pulmonary metastases, non-Hodgkin lymphoma, or mesothelioma. Some authors reported 57 cases of thoracic splenosis and four symptomatic cases in the English-language literature [5, 6]. Reviewing the MEDLINE database this is the first described case of intrathoracic splenosis in Bosnia and Herzegovina and the second symptomatic case in Europe.

CASE REPORT

A 53-year-old man was referred to our clinic because of dyspnea, frequent coughing, faster fatigue and left chest pain. He used to smoke four cigarette packs per day for 20 years and quit smoking three months before presentation. His surgical history included thoracoabdominal gunshot wound suffered 11 years previously which required an emergency splenectomy, without surgical opening of the chest. His medical history included tachycardia in the past five years. On admission, physical examination was unremarkable, and laboratory tests were normal. Chest radiographs showed oval homogeneous soft tissue opacity, measuring 4.5 × 4 cm in the left lung lobe, between first and second ribs (Figure 1). The chest CT scan revealed two well-circumscribed lobulated, nodular masses, measuring up to 2 cm in diameter in the back and the front of the left upper lobe. These nodules were not calcified. Also, a CT scan of the apical region showed subpleural fibrosis (Figure 2). In our case, technetium-99m (^{99m}Tc) scintigraphy was unavailable, so we achieved diagnosis with fine-needle aspiration, and pathohistological examination. After analysis of a sample of the biggest nodule, obtained by percutaneous aspiration biopsy and by immunochemistry analysis (vimentin+++ , LCA+++ , CD34- , S-100- , CK-), it was supposed that disease was connected to mesenchymal, cellular, predominantly lymphocytic well-vascularized, probably benign tumor, but no accurate pathologic diagnosis was made.

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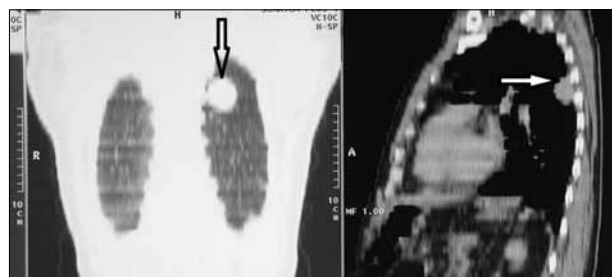


Figure 1. Chest radiograph (anterior and lateral views)

An oval smooth surface nodule of $4 \times 3.5 \times 1.5$ cm in size was removed from the apical part of the left lung lobe. In cross-section, tissue appeared soft, purple, homogenous, with adjacent several smaller oval solid nodules, also with smooth surface, size ranging from 0.3 cm to 0.7 cm, localized in the parietal pleura (Figure 3a). Histological examination of the relevant sites revealed splenic tissue with white and red pulp, lymphoid follicles, secondary germinal centers and granulocytes, surrounded by a fibrotic ring (Figure 3b). These findings were consistent with splenosis in all the relevant biopsies. No evidence of malignancy was observed.

DISCUSSION

Thoracic splenosis, first reported by Shaw and Shafi [7] in 1937, describes the autotransplantation of splenic tissue into the pleural cavity after splenectomy for traumatic or iatrogenic injury, resulting in multiple nodular implants on the left pleura. As imaging technology improved, splenosis has been noted in up to two thirds of patients after splenectomy for trauma. However, its frequency is likely underestimated because most splenic implants are asymptomatic and are only incidentally discovered during chest X-ray or CT. Abdominal splenosis is the most common type of splenosis, with most common sites of autotransplantation of the spleen being the mesentery, peritoneum, and omentum. Abdominal and pelvic splenosis are often mistaken

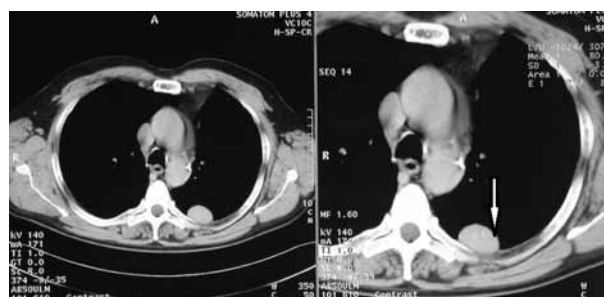


Figure 2. Thoracic computed tomography – lobulated pleural-based mass in the posterior segment of the upper lobe

for abdominal lymphoma, metastatic disease, carcinomatosis, primary renal or hepatic malignancy, adenomas, endometriosis or simple adenopathy [8]. Subcutaneous splenosis is quite rare, with less than 14 reported cases. The differential diagnosis of cutaneous splenosis includes differentiated cutaneous lymphoma, nodular Kaposi sarcoma, and subcutaneous vascular malformations. There is only one case report of intracerebral splenosis in the medical literature. Similarly, due to the spleen's anatomic location, thoracic splenosis arises almost exclusively in the left hemithorax. The most common location of thoracic splenosis occurs in the pleural cavity, and most commonly involves a diaphragmatic tear, or rarely a diaphragmatic hiatus, and small pieces of splenic tissue are displaced into the left hemithorax through the diaphragmatic opening. A second mechanism is the hematogenous spread of splenic pulp, as suggested by case reports of intrahepatic, as well as intracranial splenosis [9, 10, 11]. In fact, all cases of reported thoracic splenosis had diaphragmatic rupture as well as splenic rupture [9, 12]. In our case, spleen nodules were localized in the apex of the left lung with no evidence of them near the diaphragm. Hematogenous dissemination of splenic tissues probably occurred in this patient rather than passing through a diaphragmatic tear. Maximum amount of time that can pass between the initial trauma and the appearance of pulmonary splenosis is 40 years, and minimum is 13 years. The average interval between the initial trauma and diagnosis of thoracic

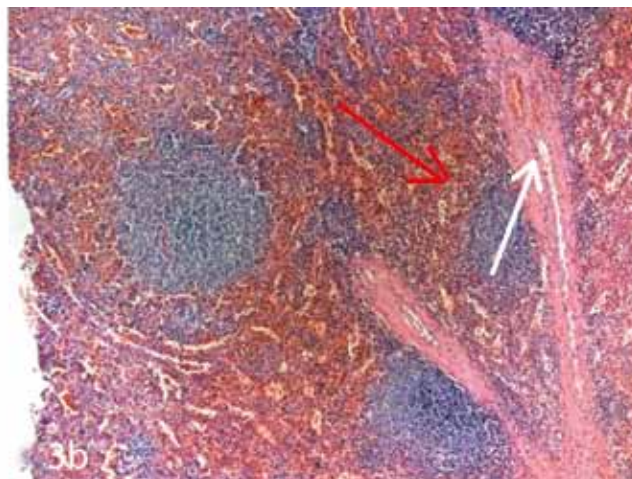


Figure 3. a) Macroscopic appearance of intrathoracic splenosis; b) microscopic appearance (haematoxylin and eosin staining; 100x): red pulp with reticular tissue and wide sinus filled with blood (red arrow); white pulp built of lymphoreticular tissue in the form of lymph nodules (Mallory bodies) with a central artery (white arrow)

splenosis is 21 years [13]. This is the first case of splenosis in which the symptoms of the disease appeared 11 years after the injury. Autotransplanted spleens differ from accessory spleens by blood supply, local perforator arteries versus splenic artery, respectively. Accessory spleens which are few in number occur near the splenopancreatic ligament and gastrosplenic ligament and have been found in up to 40% of patients undergoing autopsies. In contrast, splenosis nodules are numerous (up to 100) appearing in any location, particularly on serosal surfaces, and, though encapsulated, have no well-defined hilum. The splenic implants are sessile or pedunculated reddish blue nodules. These deposits may vary in number, they can be of any shape, and their diameter can range from a few millimeters to 12 cm. Splenosis can occur with either single or multiple nodules – upwards of 400 in some cases [14]. The diagnosis of thoracic splenosis can be established noninvasively with several diagnostic modalities: radionuclide scintigraphy or magnetic resonance imaging. Technetium-99m-labeled heat-damaged erythrocyte scanning is the preferred

method because it is significantly more specific than sulfur colloid, indium-111-labeled platelet, or ^{99m}Tc-labeled white blood cell scanning due to diminished liver uptake. Pleural lesion often requires a fine-needle aspiration if a lesion is accessible. Pathologic presentation can be misleading, with lymphocytic infiltrate misdiagnosed as lymphoma [15, 16]. If the diagnosis can be confirmed preoperatively, surgery is not indicated unless the patient is symptomatic. It is usually not necessary to remove the pulmonary nodules because the splenic tissue is slow growing, noninvasive and nonmalignant [17–20]. Four symptomatic cases of thoracic splenosis have been reported; two patients reported having hemoptysis, one patient complained of a productive cough, and a patient reported having pleuritic chest pain [21, 22]. Previously reported case of symptomatic thoracic splenosis has significant interval growth of the splenic tissue and the large size of the mass described. Our patient was indicated for operative treatment because of chest pain. We postulate that the pain in our patient was exhibited itself due to an irritation of the parietal pleura.

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Симптоматска изолована грудна спленоза 11 година после повреде трбуха – приказ болесника

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КРАТАК САДРЖАЈ

Увод Грудна спленоза представља ауотрансплантацију ткива слезине у грудни кош. Настаје због трауматског цепања слезине и истовремене повреде леве дијафрагме. Ово ретко обољење најчешће остаје неоткривено јер није праћено симптомима болести.

Приказ болесника Представљамо случај симптоматске торакалне спленозе код мушкарца старог 53 године, дугогодишњег пушача цигарета који је имао оперативно уклањање слезине због повреде трбуха и грудног коша рањавањем из ватреног оружја. Од симптома болести имао је отежано дисање, кашаљ, брзо замарање и бол у левој страни грудног коша три године пре медицинског прегледа.

Симптоми болести су упућивали на карцином плућа. На радиографији грудног коша уочава се туморска маса саграђена од неколико мањих чворова, а локализована уз плеуру у горњем режњу левог плућног крила. После хируршког уклањања туморске масе патохистолошка дијагноза је била торакална спленоза уз одсуство малигне болести.

Закључак Код пацијената са чворовима или туморским масама локализованим у левом плућном крилу и медицинском историјом трауме спленоза мора бити укључена у диференцијалну дијагнозу.

Кључне речи: симптоматска грудна спленоза; спленектомија; рањавање стомака и грудног коша ватреним оружјем

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