Pleural Nodules and Mediastinal Lymphadenopathy in a Smoker: An Unusual Case Report

Damien Desbuquoit\textsuperscript{a} Stijn Van Hecke\textsuperscript{b} Pjotr Even\textsuperscript{c} Paul M. Parizel\textsuperscript{a} Jan P. van Meerbeeck\textsuperscript{d} Annemie Snoeckx\textsuperscript{a}

\textsuperscript{a}Department of Radiology, Antwerp University Hospital and University of Antwerp, Antwerp, Belgium; \textsuperscript{b}Department of Nuclear Medicine, Antwerp University Hospital and University of Antwerp, Antwerp, Belgium; \textsuperscript{c}Department of Respiratory Medicine, Antwerp University Hospital and University of Antwerp, Antwerp, Belgium; \textsuperscript{d}Department of Thoracic Oncology, Antwerp University Hospital and University of Antwerp, Antwerp, Belgium

Keywords
Thoracic splenosis · Pleural nodules · Computed tomography · 99mTc-sulfur colloid scintigraphy · Lymphadenopathy

Abstract
The authors report a case of thoracic splenosis, which is the autotransplantation of splenic tissue into the pleural cavity. Splenosis in the chest is a rare entity and most often an incidental finding on chest computed tomography, typically showing solitary or multiple well-defined, noncalcified pleural nodules of variable size in the left hemithorax. It is important to include this benign pathology in the differential diagnosis among other, generally malignant, pleural lesions. Imaging clues to the diagnosis are absence of the spleen and/or associated rib fractures. Early identification of thoracic splenosis as a cause of pleural nodules can prevent unnecessary and risky invasive procedures, such as biopsy or surgery.
Case Presentation

A 60-year-old man with a 45-pack-year smoking history and no professional asbestos exposure presented with longstanding thoracic pain. His medical history was notable for a motor vehicle accident 11 years earlier with severe chest trauma. Physical examination showed a deformity of the left lower rib cage. A non-contrast enhanced computed tomography (CT) was performed, showing numerous well-delineated, left-sided lobulated pleural lesions (fig. 1b, c, d), besides enlarged hilar and mediastinal lymph nodes (fig. 1a, b), as well as the absence of a spleen. A tentative diagnosis of metastatic intrathoracic malignancy was advanced. Endoscopic bronchial ultrasound needle aspiration biopsy of lymph node stations 4R, 10R, 11R and 7 showed nonnecrotizing granulomas, consistent with sarcoidosis. In order to unravel the cause of the pleural lesions, a $^{99m}$Tc-sulfur colloid scintigraphy was performed, which showed elective tracer uptake in the pleural nodules (fig. 2). This confirmed the diagnosis of thoracic splenosis, already suggested by the absence of the spleen, extensive rib fracture sequelae and left-sided pleural nodules. Smoking cessation was advised, and no therapy was started for this stage I sarcoidosis.

Comments

Splenosis is a benign entity used to describe autotransplantation of splenic tissue, mainly occurring after penetrating or blunt-force trauma to the spleen and/or after splenectomy. Although previously considered a rare condition, research suggests that it occurs in up to 67% of patients with splenic rupture or splenectomy [1]. Typical locations of these ectopic implants include the mesentery, the parietal peritoneum, the greater omentum, the intestinal serosa and the abdominal surface of the diaphragm. Ectopic implantation of splenic tissue in the chest is less common and may occur when splenic rupture is accompanied by diaphragmatic laceration or through hematogenous spread. Thoracic splenosis is mostly observed as an incidental finding during imaging studies, with a mean delay of up to 21 years [2]. Symptomatic thoracic splenosis with symptoms such as pleural pain, hemoptysis and dyspnea is rare, but has been reported [3, 4]. Due to the higher rate of trauma in young men, thoracic splenosis has a male-to-female ratio of 30:8 [1]. Both visceral and parietal pleura can be affected, most often on the left side. Cases with mediastinal and parenchymal involvement have also been reported [4, 5]. CT imaging shows solitary or multiple well-defined, noncalcified pleural nodules of variable size, predominantly in the left hemithorax. After intravenous contrast administration, an enhancement pattern similar to that of the spleen can be seen [6]. Sequelae of rib fractures may be a subtle indication of previous traumatic injury to the splenic region. Pleural involvement in sarcoidosis can occur, but it is rare and manifests with pleural effusion, pneumothorax or pleural thickening. Sarcoidosis presenting with pulmonary opacities in subpleural regions can also mimic pleural disease [7]. Other differential diagnostic considerations include pleural metastases, asbestos-related pleural disease, lymphoma, localized fibrous tumor of the pleura and intrathoracic extramedullary hematopoiesis [8]. Growth of these pleural nodules should not alert the radiologist for a malignant etiology since slow growth of this ectopic tissue is a normal finding [6]. A noninvasive diagnosis can be achieved by means of a $^{99m}$Tc-sulfur colloid or $^{99m}$Tc-labeled red blood cell scintigraphy. Both these methods demonstrate elective splenic uptake, with the latter having a higher sensitivity and specificity [2]. Surgical resection of thoracic splenosis is not advocated in asymptomatic patients since it is suggested that splenic tissue im-
plants may exhibit the remaining function, and some degree of immunity can be preserved in these post-splenectomy patients.

Both thoracic splenosis and sarcoidosis were incidental findings in our patient, nor was the pain caused by either of them, but by the posttraumatic rib fracture sequelae for which he was treated with opioids. There is no known association between these entities.

Nevertheless, the combination of lymphadenopathy and pleural nodules was initially misleading for a thoracic malignancy.

Although thoracic splenosis is a rare condition, it should be considered in the differential diagnosis of pleural nodules, especially in patients with a previous history of trauma or splenectomy, exhibiting solitary or multiple well-defined, noncalcified left-sided pleural nodules. Early identification of thoracic splenosis as cause of pleural nodules can prevent unnecessary invasive procedures, such as biopsy or surgery.

**Statement of Ethics**

The authors have no ethical conflicts to disclose.

**Disclosure Statement**

The authors declare no conflicts of interest and have no disclosures.

**References**

Fig. 1. Axial non-contrast enhanced CT images in a mediastinal window setting at the level of the mediastinum demonstrate enlarged paratracheal (a) and subcarinal (b) lymph nodes (asterisk). Note the small pleural nodules in the left hemithorax (white arrows). The image of the lower thoracic region (c) shows the deformity of the rib cage and a larger well-delineated lobular pleural nodule (white arrow). The extent of the pleural abnormalities is best appreciated on the coronal reformatted image (d) showing numerous pleural nodules up to 45 mm in size. Note the absence of the spleen under the left diaphragm (arrows).
Fig. 2. SPECT/CT images after injection of 185 MBq $^{99m}$Tc-phytate (sulfur colloid scintigraphy). Axial (a) and coronal (b) fused images show physiological tracer uptake in the reticuloendothelial system of the bone marrow, liver (asterisk) as well as in the pleural nodules (white arrows), confirming the diagnosis of thoracic splenosis.