



## Case report

## Thoracic splenosis: History is the key

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## ABSTRACT

Splenosis is an acquired ectopic autotransplantation of splenic tissue; that occurs after traumatic splenic rupture and splenectomy [1]. Splenosis is a rare but benign disease, and the diagnosis can be challenging as the multiple incidentally found nodules could mimic malignancy [2].

Abdominopelvic Splenosis is thought to occur in as many as 65% of cases of splenic rupture [1]. However, Thoracic Splenosis is rare and usually involve the left parietal and visceral pleura [1,2]. Intraparenchymal lesions are less common but have been reported in cases of parenchymal and diaphragm laceration [1,2].

Taking a thorough history is of utmost importance, as these patients usually present more than two decades after the splenic traumatic rupture. The use of commonly available nuclear studies will further confirm the diagnosis [3]. This will help to avoid unnecessary procedures, like biopsies; and prevent the potential complications. We present a case of Thoracic Splenosis that highlights the importance of taking a detailed history; and the importance of using nuclear studies for the diagnosis. Further adding to its uniqueness, this case showed with multiple intraparenchymal nodules which is a less common presentation of Splenosis.

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## 1. Introduction

Diagnosing Thoracic Splenosis can be challenging [4]. However, a history of traumatic spleen rupture and splenectomy, the anatomical distribution and the dynamic behavior of the lesions on contrast-enhanced imaging studies suggest Splenosis [5]. The appropriate use of commonly available diagnostic tools can facilitate an accurate diagnosis while avoiding risky and unnecessary procedures [3]. This case highlights the importance of a detailed history and physical exam.

## 2. Case report

A 40-year-old woman presented to the pulmonary clinics at a tertiary academic medical center, for evaluation following a computer tomography (CT) scan of the chest; showing multiple pulmonary nodules. The primary physician referred her to the pulmonary clinics because the presence of those nodules raised concern for malignancy.

The CT scan was obtained during a visit to an Outside Hospital Emergency Room due to 4–5 months' history of constant progressive lower back pain. The chest CT scan showed multiple pulmonary nodules in the left lung (Fig. 1).

On evaluation in the pulmonary clinic, the patient reported improvement of her lower back pain after the treatment she received in the Emergency Room. She denied constitutional or respiratory symptoms. She had history of hyperlipidemia but was not using any medications. Otherwise, patient denied history of recurrent infections or illness. She was a lifelong nonsmoker and denied family history of lung cancer. She had a gunshot wound twenty-six years ago, requiring splenectomy; and tube thoracostomy for a left pneumothorax.

Her vital signs including pulse oximetry were normal. Physical exam was only remarkable for the scars from thoracostomy tube and bullet exit on the left side of her back.

In view of the absence of risk factors for malignancy, and the history of traumatic splenectomy, a Technetium-99m (Tc-99m) sulfur colloid nuclear scan was performed. It showed increased radiotracer uptake within pulmonary nodules in the left lower and upper lobes consistent with thoracic splenosis (Figs. 2 and 3).

In a follow up appointment, the patient was notified on the

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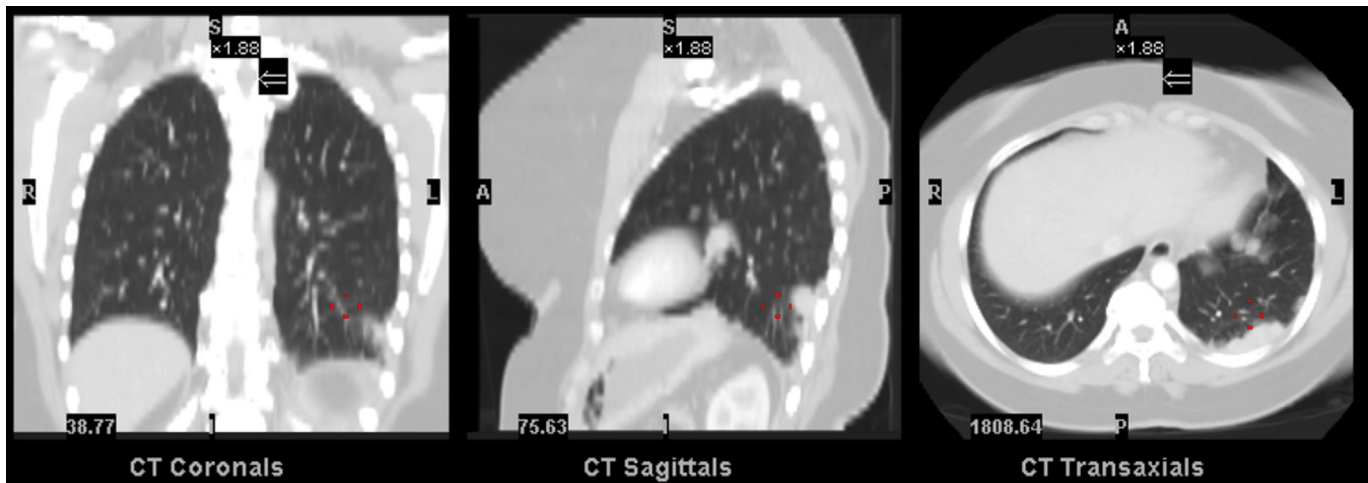


Fig. 1. Selected slices of Chest CT Scan showing multiple pulmonary nodules in the left lung. From left to right: Coronal View, Sagittal View and Transaxial View.

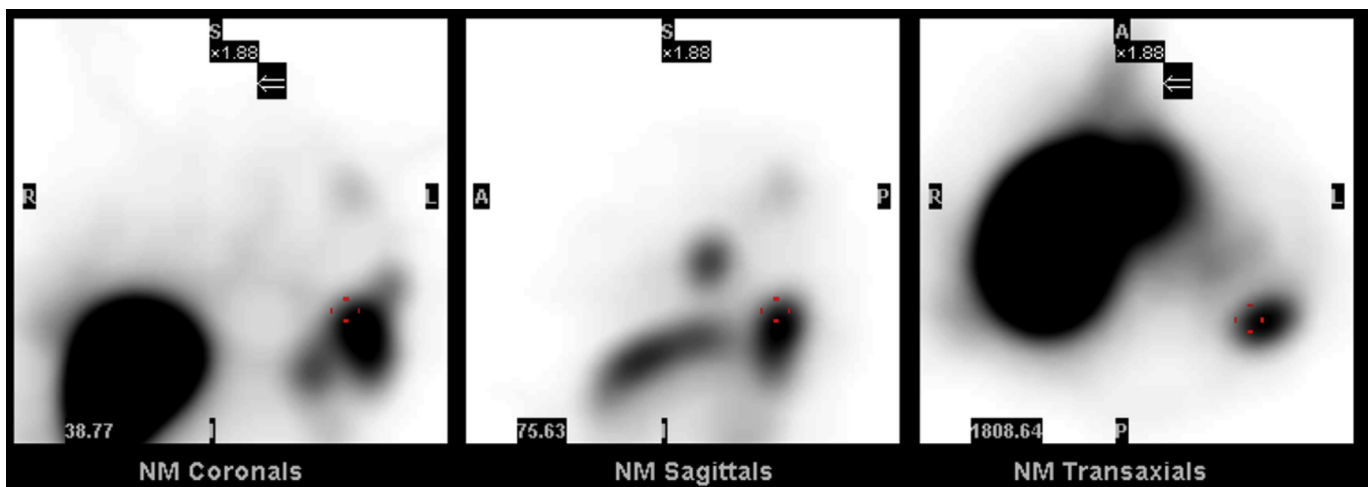


Fig. 2. Selected views of the Tc-99m sulfur colloid nuclear scan showing increased radiotracer uptake within the pulmonary nodules in the left lower and upper lobes. From left to right: Coronal View, Sagittal View and Transaxial View.

results of the nuclear scan. All her immunizations were updated and she was referred back to her primary physician.

### 3. Discussion

Splenosis is an autotransplantation of splenic tissue usually after splenic traumatic rupture and subsequent splenectomy [1]. It is presumed that spillage of the damaged splenic pulp into the adjacent cavities begins the seeding process [6]. Other mechanisms like the hematogenous spread of splenic pulp [7], and tissue growth in response to hypoxia [4], has been suggested.

It is acquired ectopic splenic tissue; usually presenting with more than one hundred nodules with distorted architecture and very variable histology. The nodules derive their blood supply from surrounding tissues and vessels [1]. Kim et al. 2010, described these nodules as having a gross appearance of a hemangioma containing blood [2].

They are most commonly found in the abdomen or pelvis, with seeding of peritoneum, omentum or mesentery [8]. Abdominopelvic splenosis, occurs in up to 65% of splenic rupture cases [1]. Thoracic splenosis occurs much less frequently, in possibly 18% of post splenic rupture cases [1]. It is associated with a history of

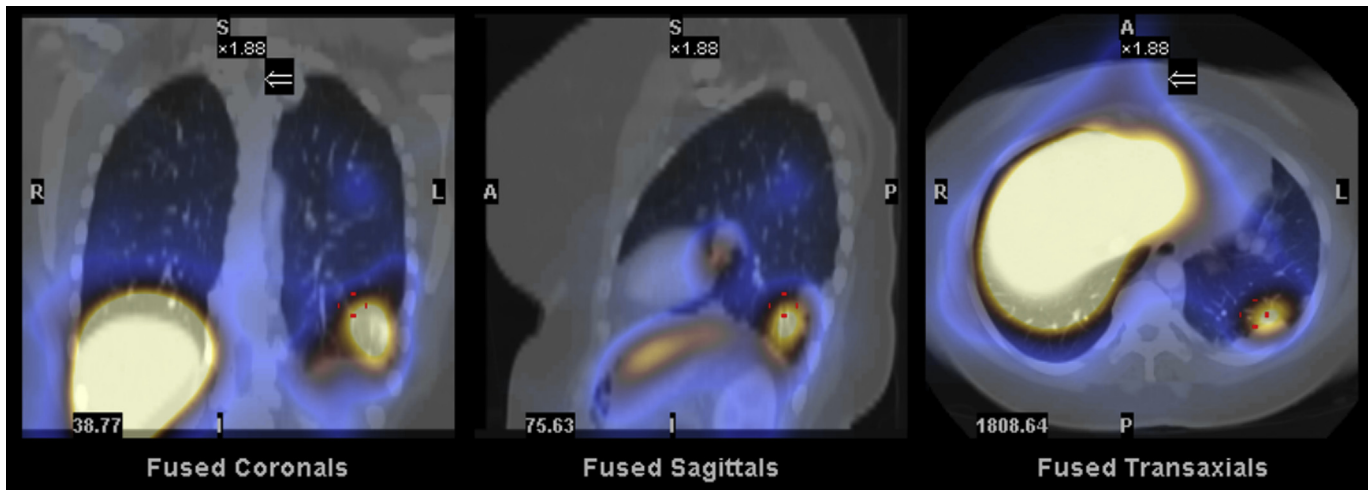
traumatic rupture of the spleen and diaphragm [9,10]. The nodules are almost exclusively in the left hemithorax commonly accompanied by abdominal splenosis [2]. They develop from pleura, although intraparenchymal lesions has been reported with diaphragmatic and parenchymal laceration [1,2].

Splenosis is an incidental finding, as patients are usually asymptomatic [3]. Pleurisy and/or Hemoptysis are rarely reported [1].

The diagnosis is challenging since there is an average interval between initial trauma and diagnosis of approximately twenty-one years [2]. Pleural-based nodules may be mistaken for an intrathoracic malignancy, prompting needle or Video-assisted thoracic surgery (VATS) biopsy [11].

The gold standard for diagnosis is Technetium-99m (Tc-99m) heat-damaged erythrocyte nuclear study; where the erythrocytes are sequestered, and phagocytosed in the spleen, increasing the uptake compared to the liver [12,13]. It has the highest specificity and sensitivity. Tc-99m sulfur colloid and indium-111-labeled platelets are alternative examinations, although less sensitive. The Tc-99m white blood cell scan is the least specific study, as it is mainly used for the diagnosis of osteomyelitis [13].

Ferumoxide Magnetic Resonance Imaging (MRI) provides a



**Fig. 3.** Selected views of the Fused image showing increased radiotracer uptake within the pulmonary nodules in the left lower and upper lobes. From left to right: Coronal View, Sagittal View and Transaxial View.

higher spatial resolution, since superparamagnetic iron oxide are taken up by the reticuloendothelial system. More research is needed to directly compared nuclear studies to Ferumoxide MRI in diagnosing splenosis [13].

Some cases may be difficult to diagnose and require a thoracotomy. CT-guided biopsies and needle aspirations have returned inconclusive or misleading in reported cases [13]. Furthermore, the increased risk of bleeding outweigh the benefit of percutaneous biopsy [2]. Peripheral smears for Howell-Jolly bodies or pitted erythrocytes is neither sensitive or specific [14].

Splenosis is a benign condition [11]. The splenic tissue is slow growing, non-invasive [15] and surgical removal of the nodules is generally inadvisable [10]. The excision of implants could trigger serious bleeding and damage of surrounding organs [16]. Rarely video-assisted thoracoscopic surgery may be required for removal of implants in symptomatic patient with hemoptysis, cough or pleuritic chest pain [15].

The protective role of splenosis is controversial, although patients have been reported to have a decreased rate of post-splenectomy sepsis [13]. However, it is still associated with reduced immune function as the residual volume and function is insufficient to confer protection against overwhelming post-splenectomy infection. Therefore, there is need for immunization and early prophylactic measures [3].

## References

- [1] A.H. O-Yurvati, J.B. Thompson, T.N. Woods, Thoracic splenosis more than 40 years after thoracoabdominal trauma, *J. Am. Osteopath Assoc.* 113 (11) (2013 Nov) 853–856.
- [2] K. Kim, H.J. Choi, Y.M. Kim, W.J. Kwon, W.C. Lee, J.H. Suh, Thoracic splenosis: a case report and the importance of clinical history, *J. Korean Med. Sci.* 25 (2) (2010 Feb) 299–303.
- [3] L. Tulinský, P. Ilnát, M. Mitták, P. Guňková, P. Zonča, Intrathoracic splenosis - lesson learned: a case report, *J. Cardiothorac. Surg.* 11 (1) (2016 Apr) 72.
- [4] C.M. Kwok, Y.T. Chen, H.T. Lin, Portal vein entrance of splenic erythrocytic progenitor cells and local hypoxia of liver, two events cause intrahepatic splenosis, *Med. Hypotheses* 67 (2006) 1330–1332.
- [5] I. Sánchez-Paniagua, S. Baleato-González, R. García-Figueiras, Splenosis: non-invasive diagnosis of a great mimicker, *RevEsp Enferm. Dig.* 108 (1) (2016 Jan) 40–41.
- [6] R.D. Fremont, T.W. Rice, Splenosis a review, *South. Med. J.* 6 (2007 June 100) 589–593.
- [7] C.J. Yeh, W.Y. Chuang, T.T. Kuo, Case report: unusual subcutaneous splenosis occurring in a gunshot wound scar: pathology and immunohistochemical identification, *Pathol. Int.* 56 (2006) 336–339.
- [8] J.P. Normand, M. Rioux, M. Dumont, et al., Thoracic splenosis after blunt trauma: frequency and imaging findings, *Am. J. Roentgenol.* 161 (1993) 739–741.
- [9] J.S. Klair, C. Duvoor, N. Meena, A rare benign intrathoracic mass in a patient with history of rocket explosion, *Respir. Med. Case Rep.* 14 (2014) 4–6.
- [10] S. Fukuhara, S. Tyagi, J. Yun, M. Karpeh, A. Reyes, Intrathoracic splenosis presenting as persistent chest pain, *J. Cardiothorac. Surg.* 7 (2012) 84.
- [11] M. Remtulla, N.E. Drury, N.A. Kaushal, S.E. Trotter, M.S. Kalkat, Thoracic splenosis masquerading as advanced lung cancer, *Thorax* 0 (2016 Sept) 1–2.
- [12] J.N. Yammine, A. Yatim, A. Barbari, Radionuclide imaging in thoracic splenosis and a review of the literature, *Clin. Nucl. Med.* 28 (2) (2003 Feb) 121–123.
- [13] U.F. Malik, M.R. Martin, R. Patel, A. Mahmoud, Parenchymal thoracic splenosis: history and nuclear imaging without invasive procedures may provide diagnosis, *J. Clin. Med. Res.* 2 (4) (2010 Aug 18) 180–184.
- [14] N.T. Connell, A.M. Brunner, C.A. Kerr, F.J. Schiffman, Splenosis and sepsis: the born-again spleen provides poor protection, *Virulence* 2 (1) (2011 Feb) 4–11.
- [15] W. Bugiantella, F. Crusco, N. Avenia, R. Fabio, Thoracic splenosis. Report of a case and review of the diagnostic workup, *Ann. Ital. Chir.* 87 (2016 Jul 29).
- [16] A. Matthews, M. Chesser, J. Mand, A. Thomas, A growth opportunity: thoracic Splenosis, *Am. J. Med.* 130 (4) (2017 Apr) 420–422.