International Journal of Innovative Research in Medical Science (IJIRMS)
Volume 10, Issue 01, January 2025,
https://doi.org/10.23958/ijirms/vol10-i01/2017

Case Report

Thoracic Splenosis: The Luckiest Nodule You Can Have

Marta Mello Vieira *, Sara Barbosa, Mariana Morais, Leonor Soares, Maria do Carmo Fevereiro

Unidade Funcional de Medicina 1, Hospital de São José - Unidade Local de Saúde de São José, 1150-199, Lisbon, Portugal.

*Corresponding author: Marta Mello Vieira; *marta.vieira@ulssjose.min-saude.pt*

Received 17 December 2024; Accepted 02 January 2025; Published 05 January 2025

Abstract

Thoracic splenosis is characterised by the autotransplantation of splenic tissue into the thoracic cavity, following splenic and diaphragmatic rupture. It is usually asymptomatic and diagnosed incidentally.

The authors present the case of a 61-year-old woman, with a history of a car accident 36 years prior which resulted in splenectomy. The patient presented to the emergency department with a syncytial respiratory virus infection, which led to the identification on computed tomography of extensive nodular thickening of the left peripheric pleura (11cm extension, 2,9cm thickness), suggestive of neoplasia. She underwent a percutaneous pleural biopsy, followed by video-assisted thoracoscopy, which revealed lymphoid tissue with germinal centres and abundant lipid-laden CD68+ macrophages. A positron emission tomography with 18F-FDG identified a low metabolic signal on the left pleura. Follow-up chest computed tomography demonstrated the stability of the nodular lesions. After a multi-disciplinary discussion, given the patient's medical history, the hypothesis of thoracic splenosis was considered the most likely diagnosis.

This case brings our attention to this rare entity, highlighting the importance of a detailed clinical history, a persistent search for a diagnosis and a multi-disciplinary approach.

Keywords: ***splenosis; thoracic splenosis; pleural nodule; benign nodule; splenic rupture***

Introduction

Thoracic splenosis is the heterotopic autotransplantation of splenic tissue inside the thoracic cavity after simultaneous splenic and diaphragmatic rupture [1-7]. It is a rare, benign condition [1,4,6-7], usually asymptomatic and incidentally diagnosed [1-5,7-9], on average 21 years after the provoking event [1,10]. Thoracic nodules can point to different aetiologies, such as primary malignancy, metastatic disease, and infection [1,2,6,10], which presents a diagnostic challenge. Nuclear imaging study with heat-damaged erythrocytes tagged with technetium-99 or nodule biopsy, together with a careful patient history review leads to confirmation of diagnosis [1,3-5,9,10,11].

We describe a case of thoracic splenosis presenting 36 years after a car accident with splenic trauma and splenectomy, which was diagnosed after a respiratory viral infection led to thorax imaging.

Case report

A 61-year-old woman, with a known medical history of type 2 diabetes mellitus, dyslipidaemia, obesity, hypothyroidism, depressive syndrome and a car accident 36 years prior with polytrauma, from which resulted fracture of multiple costal arches, need of left thoracic drainage and splenectomy. The patient arrived at the emergency department with a 9-day history of cough with phlegm, dyspnoea, asthenia and myalgias. She had been medicated with azithromycin with no improvement. On physical examination, she was eupnoeic with normal oxygen saturation and presented wheezing on pulmonary auscultation. Blood tests revealed an anaemia with haemoglobin 11g/dL, normal white blood cell count of 10860 leucocytes/L and 5070 neutrophils/L as well as normal C-reactive protein of 7,8mg/L and a chest X-ray showed a nodule on the lateral region of the left lung (**see Figure 1**). A chest computed tomography (CT) was then performed which confirmed extensive nodular thickening of the left peripheric pleura (11cm anteroposterior extension, 2,9cm of maximal thickness), suggestive of mesothelioma or lung neoplasia (**see Figure 2**); these lesions were in proximity with consolidated fractures of several costal arches. After a respiratory virus search, she was diagnosed with a syncytial respiratory virus infection. She was hospitalised, where initial studies were performed: a mammary ultrasound and an abdomen-pelvic CT, showing no relevant changes, and a search of acid-fast-bacilli on gastric aspirate which was negative.

Due to clinical improvement, the patient was discharged to proceed further investigation in an ambulatory context. She was submitted to a percutaneous pleural biopsy guided by CT, which revealed lymphoid tissue with abundant macrophages and the absence of cancer cells. Follow-up chest CTs 2 and 5 months after discharge showed stability of the nodular pleural lesions and two retro-oesophageal masses (dimensions 2.7 x 2.4cm and 1.6 x 0.9cm) already present on the first CT, also stable and likely with the same aetiology; in the spleen orthotopic position the was a dysmorphic-looking mass suggestive of residual splenic tissue. A positron

emission tomography with 18F-FDG only identified a low metabolic signal on the left pleura (**see Figure 3**). The patient was re-biopsied by video-assisted thoracoscopy, revealing fragments of skeletal muscle and lymphoid tissue with germinative centres and abundant lipid-laden macrophages CD68+. Clinically, on follow-up the patient presented occasional episodes of exertional dyspnoea, motivating a transthoracic echocardiogram which identified mild mitral and tricuspid regurgitation and a thickening of the walls of the left ventricle with normal systolic function. After a multi-disciplinary discussion, the hypothesis of thoracic splenosis was considered the most likely diagnosis, given the patient's medical history. In the absence of other symptomatology, it was decided to take a conservative approach with regular appointments and imaging revaluation.

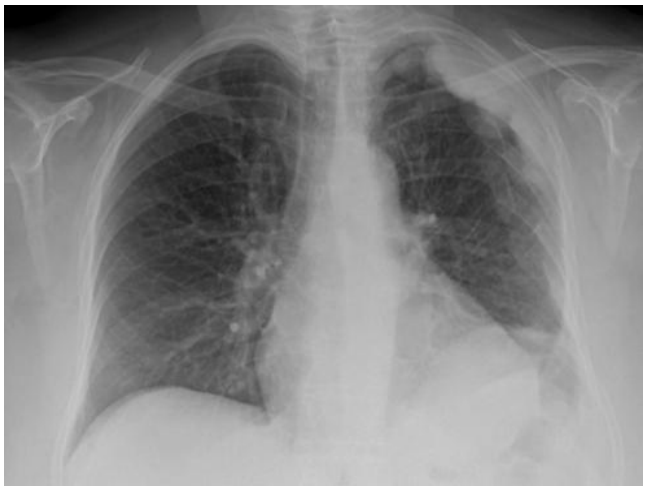

Figure 1: Thorax radiography showing a pleural nodularity on the left upper hemithorax.

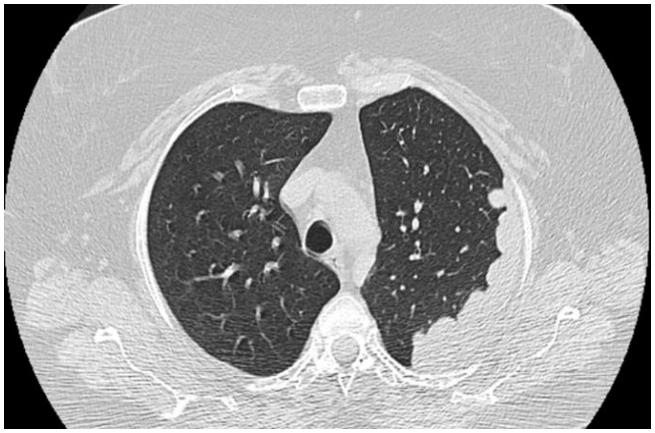

Figure 2: Thorax computed tomography, where extensive nodular thickening of the left peripheric pleura can be observed.

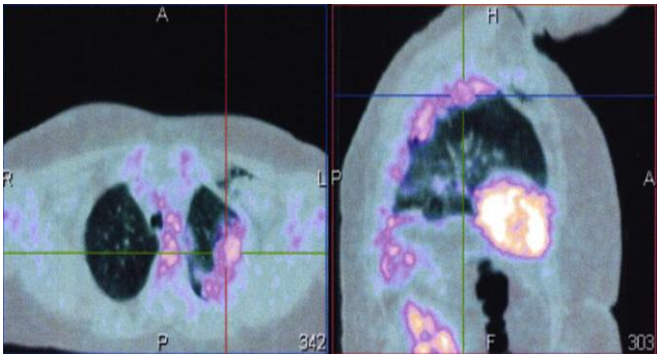

Figure 3: Positron emission tomography with 18F-FDG identified a low metabolic signal on the left pleura.

Discussion

Splenosis describes the presence of splenic tissue in other anatomic areas of the body after splenic rupture [1-8,10,11]. It is thought that the splenic tissue is implanted into adjacent cavities [1-3,6,8,10,11,13], although haematogenous spread has also been hypothesised in cases of splenosis in more distant locations (for example, intrahepatic and intracranial splenosis) [1,3,12]. The splenic tissue derives its blood supply from surrounding tissues [1,4,8] and, with the help of adult spleen reticulum cells, develops splenic nodules with functional activity [1-3,8,10,13]. Most frequently the splenic tissue is found in the abdominal cavity [1,2,4,5,10,11], but, if there is concomitant diaphragmatic rupture, intrathoracic splenosis can occur, as first described by Shaw and Shafi in 1937 [5,6,8,9]. Although rarely diagnosed, it is estimated that splenosis occurs in 76% of patients after accidental or iatrogenic splenic rupture and intra-thoracic splenosis in 18% of patients with accompanying diaphragmatic injury [3,4,6,8-10] with the time between the inciting event and the diagnosis ranging from 3 to 50 years with an average of 21 years [1,10]. All cases described up until this moment refer to lesions localised in the left hemithorax and pleura-based [1,2,5-8,10,12,13]; the majority are characterized by multiple nodules (75%), while only 25% of cases showing solitary pleural nodules [1,2]. There is a male predominance of 3:1, which has been attributed to the higher rate of trauma in young men [3], notably gunshot wounds, even though there has been a rising number of cases described after motor vehicle accidents in more recent years [9]. In the case presented in this article, the diagnosis was made 36 years after a car accident with polytrauma, fracture of multiple costal arches, need of left thoracic drainage and splenectomy. Although we do not have information regarding a diaphragmatic injury, given the rib fractures and thoracic drainage, we can assume that there was probably a diaphragmatic disruption, allowing the migration of splenic tissue onto the thoracic cavity.

Pleural nodules are usually ≤3 cm in diameter, round, smooth or sessile [3,9,10,13] and, in general, do not cause symptoms [1-3,5,7,8-10,11] which explains the long time between the lesion and the diagnosis, in contrast with abdominal splenosis which more commonly presents with abdominal pain or intestinal obstruction. There are a few cases described in the literature of symptomatic presentation typically related to larger lesions, with dyspnoea, haemoptysis and thoracalgia [3-6,8-10,11], although there is limited evidence if the symptomatology was caused by the splenic nodules, since the patients presented with other comorbidities, such as heart failure, pulmonary disease and obesity hypoventilation syndrome [4]. The patient in our case presented with a history of cough with phlegm, dyspnoea, asthenia and myalgias most likely related to a syncytial respiratory virus infection, which led to thorax imaging exams and the incidental discovery of pleural nodules. She substantially improved with supportive treatment only, with the possibility of discharge, although she maintained recurrent episodes of exertional dyspnoea, which were attributed to her comorbidities and not the thoracic splenosis.

Diagnosis of thoracic splenosis is challenging because the nodules can mimic other conditions, including primary malignancy or secondary lesions, infection and reactive lymph nodes [1,2,6,7,10]. Nodules are usually revealed by radiography, CT or ultrasound, but none of these exams can determine their origin [1,2,5,13]. On intravenous contrast-enhanced CT, nodules have the same attenuation as the normal splenic tissue [1,3,5,10,13], and on magnetic resonance imaging (MRI) they also have a similar intensity to normal splenic tissue, being isointense to muscle on T1 and slightly hyperintense on T2 [1,3,11,13]. MRI with intravenous superparamagnetic iron oxide has been used due to this contrast's

specificity for the cells of the reticuloendothelial system of the liver and spleen [1,3]. Nuclear scintigraphy is an important diagnostic tool, with the use of technetium-99m-labelled sulphur colloid or heat-damaged red blood cells tagged with technetium-99m, which will be taken by the ectopic splenic tissue [1-5,9-11]. On single photon emission computed tomography (SPECT), the splenic tissue will coincide with areas of radionuclide uptake [3,10].

Classically, a biopsy was considered essential for a definite diagnosis, with transthoracic fine needle aspiration cytology being rarely useful, leading to surgery or video-assisted thoracic surgery to obtain a tissue sample [3,4,11]. On histopathology, splenic implants appear with a distorted architecture, showing lymphoid follicles, sometimes with areas of red pulp and white pulp, but lacking trabecular structure and with a poorly formed capsule [3,6,9,10]. More recently, however, nuclear scintigraphy using heat-damaged red blood cells tagged with technetium-99m is being considered the method of choice for diagnosis as it is non-invasive and carries a minor risk of complications [1-5,9-11]. A history of traumatic spleen rupture is also necessary for the diagnosis [3,5,6].

In the case described, the diagnostic procedures began after thorax radiography and CT identified pleural nodules suggestive of mesothelioma. Subsequent CTs did not show any progression, which could have been seen, given that the splenic tissue is active and can grow. An initial percutaneous pleural biopsy identified lymphoid tissue with abundant macrophages, leading to a video-assisted thoracoscopy which showed, once again, lymphoid tissue with germinative centres and abundant foam cells. Although this is not a classic description of splenic tissue, in splenosis the architecture of splenic tissue is distorted, revealing mainly lymphoid follicles which is compatible with the findings. The positron emission tomography with 18F-FDG revealed a low metabolic signal on the left pleura, which is also consistent with the diagnostic suspicion. Nuclear scintigraphy was not performed which might have led to a more precocious diagnosis and avoided the second biopsy.

Asymptomatic patients do not require specific treatment as it is a benign condition, and the risks of surgery outweigh the benefits [1,2,4,6-7]. It has been conjectured that the splenic tissue might even confer some protection in patients submitted to splenectomy [3,5,8,10], although some authors consider that the amount of splenic tissue is insufficient to have this effect [3]. Excision of splenic implants can be considered if the patient is symptomatic, or if the diagnosis is not confirmed [3,7,11].

Conclusions

Though it can mimic several conditions, including malignancy, thoracic splenosis is a benign disorder. This case demonstrates the importance of a careful and detailed clinical history to reach a correct diagnosis. The rarity of this entity, the fact that it occurs several years after a traumatic event, together with the long list of differential diagnoses, leads to a diagnostic challenge, normally with unnecessary invasive techniques, which was the case in our patient. In agreement with known literature, our patient's symptomatology also couldn't be explained by the thoracic masses, remaining a mostly asymptomatic condition with no need for further treatment.

Decelerations

Ethics approval and consent to participate

Consent was given by the patient for the writing of this article.

List of abbreviations

CT: Computed tomography

MRI: Magnetic resonance imaging

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Funding Statement

The authors were not funded by any institution.

Authors' contributions

MMV was involved in patient care, collected and analysed the patient data and wrote the manuscript. SB was involved in patient care and was a major contributor in reviewing the manuscript. MM and MCF were major contributors in reviewing the manuscript. All authors read and approved the final manuscript.

References

[1] S. T. Lake, P. T. Johnson, S. Kawamoto et al, "CT of Splenosis: Patterns and Pitfalls," AJR, vol. 199, pp. W686–W693, 2012.

[2] J. M. Kim, S. M. Lee, J. Choi et al, "Two Rare Cases of Intrathoracic Splenosis and Subcutaneous Splenosis: Spleen Scintigraphy Avoided the Need for Invasive Procedures,", Nucl Med Mol Imaging, vol. 50, pp. 76–79, 2016.

[3] M. C. F. de Paula, D. L. Escuissato, L. C. Belém et al, "Focal pleural tumorlike conditions: Nodules and masses beyond mesotheliomas and metastasis", Respiratory Medicine, vol. 109, pp. 1235-1243, 2015.

[4] O. Ajala, L. Yoe, T. Decatur et al, "Thoracic Splenosis in the Setting of Abdominal Trauma", Cureus, vol. 14, no. 8, Article ID: e27851, 2022.

[5] S. Madjar and D. Weissberg, "Thoracic splenosis", Thorax, vol. 49, pp. 1020-1022, 1994.

[6] S. Tian, Y. Cai and W. Tian, "Thoracic splenosis mimicking Castleman's disease: A case report and review of the literature," Asian Journal of Surgery, vol. 45, pp. 1480-1481, 2022.

[7] M. J. Araújo, J. Cunha and L. Ferreira, "Thoracic Splenosis, a Challenging Diagnosis", Arch Bronconeumol., vol. 57, no. 5, pp. 369, 2021

[8] F. Le Bars, R. Pascot, C. Ricordel et al, "Thoracic splenosis: Case report of a symptomatic case", Chinese Journal of Traumatology, vol. 23, pp. 185-186, 2020.

[9] N. Harb, J. Fattore, M. Saththianathan et al, "Thoracic splenosis: Precision medicine can prevent thoracic surgery", Respirology Case Reports, vol. 12, Article ID: e70067, 2014.

[10] T. Ullah, S. Chauhan, J. Friedman et al, "Calcification in Thoracic Splenosis", Case Reports in Pulmonology, vol. 2022, Article ID 9538355, 2022.

[11] N. Arish, A. Rokach, A. Jarjou'i et al, "Thoracic Splenosis: The Importance of History Taking", Imaj, vol. 24, pp. 61-63, 2022.

[12] B. Jankharia, "Thoracic splenosis: A curious case of thoracic masses", Natl Med J India, vol. 35, pp. 93-94, 2022.

[13] F. M. C. Pessôa, A. S. A. de Melo, A. S. Souza et al, "Applications of Magnetic Resonance Imaging of the Thorax in Pleural Diseases: A State-of-the-Art Review," Lung, vol. 194, pp. 501-509, 2016.