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Post-traumatic Intrathoracic and Subcutaneous Splenosis

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Abstract Splenosis is a benign condition that could found incidentally and usually asymptomatic. It is associated transplantation of splenic tissue to non-splenic organs after splenic rupture. This process could be triggered after trauma or surgery.The mechanism behind splenic cell autotransplantation begins with the splenic rupture, either from trauma or surgical removal. Seeding of splenic tissue could occur various locations. Subcutaneous splenosis is an extremely rare form of splenosis, mostly observed in abdominal surgical scars. In the presence of coexisting injury to the diaphragm intrathoracic transplantation can occur emerging as single or multiple pleural-based masses. In our case report we describe splenosis combined intrathoracic and subcutaneous involvement, as well as a comprehensive review of the literature. A 64-year-old man, with a past medical history of splenectomy two years ago came to clinic for repairing abdominal hernia. During abdominal CT scan we incidentally found subcutaneous and pleural seeding of splenic tissue.

Keywords: *Splenic trauma, Splenosis, Subcutaneous nodules, Pleural splenosis, Diffusion weighted imaging (DWI), Hematoxylin and Eosin (H&E) staining*

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1. Introduction

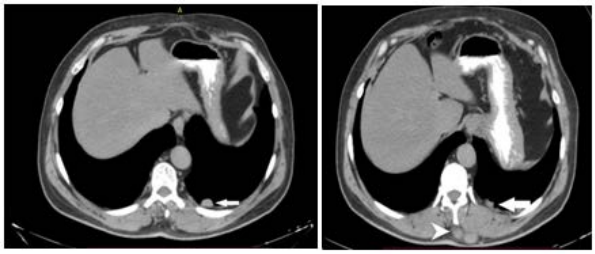
Splenosis is a term initially used by Buchbinder and Lipkoff in 1939 to describe the heterotopic transplantation of splenic tissue as a consequence of splenic trauma or surgery [1]. Splenosis is a benign and rare acquired condition in which auto-transplantation of focal ectopic splenic tissue implants occurs in heterotopic location throughout different anatomic compartments, such as the peritoneal and pelvic cavities, even the thoracic cavity and subcutaneous tissue following splenectomy. The true incidence of this condition is unknown because splenosis is usually an incidental finding at surgery, autopsy, or imaging. However, splenosis has been reported in up to 67% of patients with a history of splenic trauma [2].

We report a case of combined intrathoracic and subcutaneous splenosis in a 64-year-old male who had splenectomy after traumatic splenic rupture two years ago during the war. Our study includes clinical presentation, Magnetic resonance imaging (MRI), and computed tomography (CT) findings, confirmed by the histological study.

2. Case Report

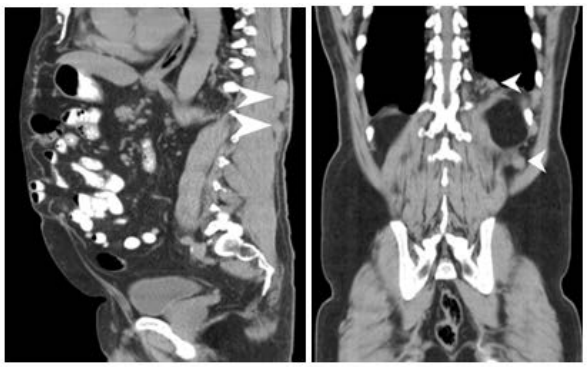
A 64-year-old man was referred to our clinic with complaints of abdominal pain and swelling. The surgeon detected an abdominal wall hernia on physical examination and ordered a CT scan to identify the exact location and assess the inside of the hernia in depth. The past medical history of the patient consists of splenectomy and left nephrectomy two years ago after gunshot trauma during war. On non-enhanced CT was detected incidental multiple left pleural-based and subcutaneous nodular masses at the site of the abdominal incision (Figure 1, Figure 2).

Thoracic and subcutaneous splenosis were suspected due to his history of abdominal trauma and the nature of the lesions on CT imaging but there was substantial concern for malignant tumor or pleural metastasis. For further diagnosis was ordered contrast enhanced abdominal MRI (Figure 3, Figure 4) and histological examination (Figure 5, Figure 6) of subcutaneous nodular mass.



A B

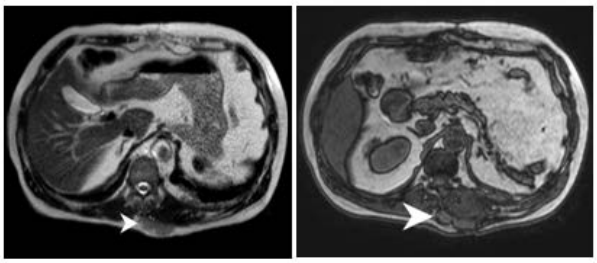
Figure 1. Non-enhanced abdominal CT Axial images showing a rounded pleural-based soft tissue nodule (A, B. white arrows) and multiple solid nodular subcutaneous lesions at the left upper abdominal wall (B. white arrowhead)



C D

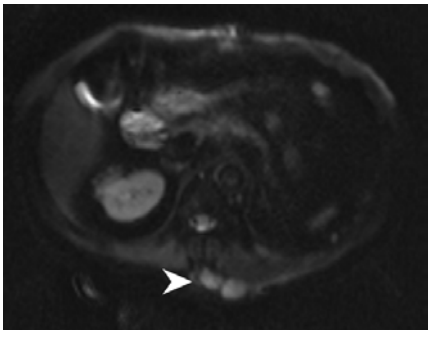
Figure 2. Non-enhanced abdominal CT Sagital (C) and Coronal (D) images

C. multiple solid nodular subcutaneous lesions at the left upper abdominal wall (white arrowhead). D. multiple subcutaneous and left pleural-based nodular lesions (white arrowhead).



A B

Figure 3. Abdominal MR Axial T2 (A) and T1 (B) imaging: Multiple subcutaneous nodular lesions are isointense with paravertebral muscles on T1WI, hyperintense on T2WI (white arrowheads)



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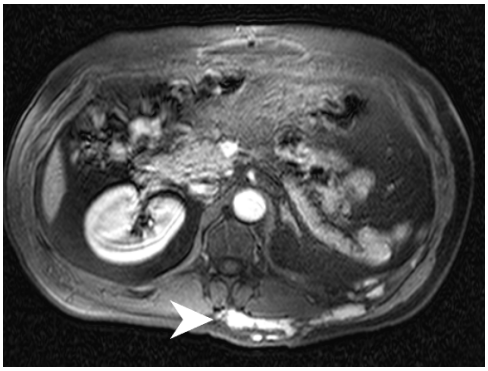

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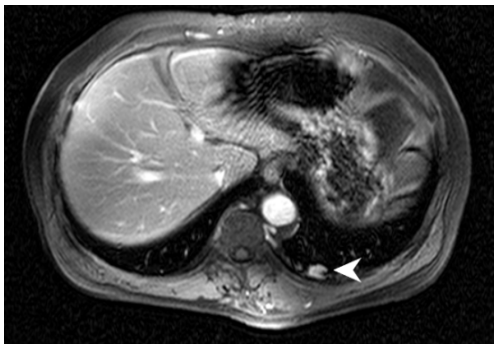

E

Figure 4. Abdominal MR Axial DWI (C) and T1 post-contrast Fat-sat images (D, E): subcutaneous nodular lesions are showing high signal on diffusion-weighted images (C. white arrowhead), homogeneously enhanced multiple subcutaneous nodular lesions (D. white arrowhead) and left pleural-based nodular lesion (E. white arrowhead) after contrast administration

Histological examination of excised subcutaneous nodular lesion confirmed the presence of splenic tissue with white and red pulp, and no evidence of malignancy (Figure 5, Figure 6). The connective tissue also appeared to contain blood vessels adjacent to splenic red pulp (Figure 5,Figure 6). The red pulp consisted of a complex network of venous sinuses and cords of Billroth, which contain most of the splenic macrophages.

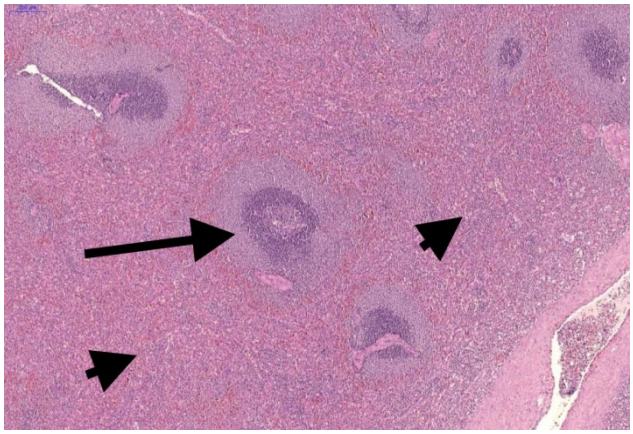

Figure 5. Hematoxylin & Eosin stain at x40 magnification. Splenic parenchyma is composed of the red pulp (arrow heads) and white pulp (arrow). White pulp with primary and secondary follicles composed of mature B-lymphocytes. Red pulp with sinuses, vasculature, and cords of Billroth

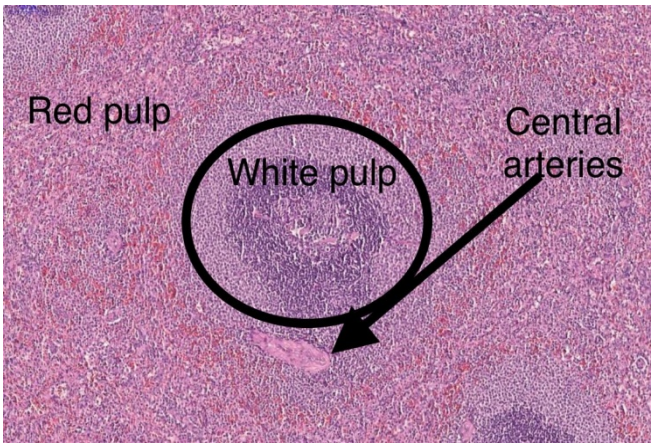

Figure 6. Hematoxylin & Eosin stain at x200 magnification. The splenic tissue consists of red pulp, white pulp, and central arteries. Thick connective tissue containing blood vessels next to the red pulp of the spleen

These findings supported the diagnosis of thoracic and subcutaneous splenosis. The nodules were not further excised, as no further workup is necessary, once splenosis is confirmed, unless the patient is symptomatic.

3. Discussion

Splenosis is an acquired condition and is defined as the autotransplantation of splenic tissue following abdominal or splenic trauma or splenectomy, displacing fragmented splenic tissues which may subsequently regrow at implanted sites by acquiring a vascular supply. The exact pathogenesis of splenosis is unknown but is suggested to be related to mechanical trauma and splenic rupture releasing splenic pulp into the surrounding tissues [3]. Splenic implants are usually multiple and small [6], with a distorted architecture with no hilum, poorly formed capsules, and variable in shape and size [8]. They receive blood supply from nearby arteries at the site of implantation that can occur in any part of the body: they may be localized in the peritoneum [8,12], in the thorax [14], in the pelvis [17,18], in the subcutaneous tissue [19,20] and even cerebrum [11].

İn most cases people with splenosis have no symptoms, so the splenosis is discovered by chance through screening or in the process of diagnosing another disease. Some people experience symptoms, such as abdominal pain, intestinal obstruction [15], hemorrhage [21], or hydronephrosis [23]. Tissue infarction due to limited blood supply can cause symptomatic splenosis.

In the presence of coexisting diaphragm abnormalities or injury intrathoracic implantation may occur leading to one or more pleural-based splenic nodules [4]. Thoracic splenosis was first discovered in an autopsy of a 25-year old male in 1896 and later in six autopsies reported by Shaw and Shafi in 1937 [7]. Due to the location of the spleen and anatomic boundaries intrathoracic autotransplantation almost always occurs into the left hemithorax. In addition to subpleural ectopic implants there have also been pulmonary intraparenchymal implants in patients with concurrent pulmonary laceration [5]. Thoracic splenosis is generally an asymptomatic condition but there have been thoracic cases presenting with pleurisy [9] and hemoptysis [16].

Subcutaneous splenosis is rare and is rare and is always associated with the scar site of a surgical incision or a traumatic wound [13]. In a 2006 review [20] of 11 cases of subcutaneous splenosis, investigators described splenic implants in old abdominal scars in eight cases and in exit gunshot wounds in three cases. This rare finding may be confused with lymphoma, cutaneous lymphoid hyperplasia, Kaposi sarcoma, or subcutaneous vascular malformations [3].

In patients with history highly suggestive of splenosis and without risk factors or symptoms indicative of malignancy diagnosis can be obtained nonsurgically using Computed tomography (CT), Magnetic Resonance Imaging (MRI) and radionucleotide scintigraphy [22,24,25].

On unenhanced and contrast-enhanced CT, the masses are similar in attenuation to the expected appearance of normal splenic tissue. On MRI, the intensity and enhancement of the splenic implants on T1- and T2-weighted images and on dynamic MR images appear to be identical to a normal spleen [8].

Tc-99m sulfur colloid scan selective splenic imaging allowing the noninvasive diagnosis of this entity avoiding more aggressive diagnostic techniques such as fine needle aspiration or laparotomy.The additional use of hybrid imaging (combining SPECT and CT) improves specificity through a more precise localization of ectopic splenic tissue [24].

When the diagnosis remains unclear, further biopsy or endoscopic excision is recommended [10]. The autotransplanted splenic tissue may exhibit red and white pulp that appears histologically normal by routine methods, and a well-developed capsule and trabeculae may form.

Because splenosis is generally asymptomatic, surgical excision of ectopic splenic nodules is often unnecessary and may subject patients to unnecessary surgical risks.

4. Conclusion

To conclude, we present a extremely rare case of post-traumatic combined thoracic and subcutaneous splenosis. It represents a diagnostic challenge that may often lead to confusion and cause the patient to be subjected to needless surgical operations and follow-up for metastatic disease. In all patients with a history of a splenic rupture or splenectomy, subcutaneous splenosis should be considered in the differential diagnosis of a subcutaneous nodule, especially when it occurs in a surgical or gunshot wound scar, and also intrathoracic splenosis should be considered in the differential diagnosis of left pleural nodule. Immunohistochemistry is useful in order to confirm the diagnosis.

Conflict of Interest

The authors state that there is no conflict of interest.

Abbreviations

CT- Computed Tomography
MRI- Magnetic Resonance Imaging
Hematoxylin and Eosin (H&E) staining

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