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Isolated Splenic Peliosis: Case Report

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Abstract **Case Report**

Peliosis is a condition characterized by the appearance of multiple cystic cavities filled with blood in the parenchyma of solid organs. Although the condition most commonly affects the liver, it can also affect the spleen, lungs, bone marrow, kidneys, and lymph nodes. The case reported here concerns a 56-year-old man with recurrent left hypochondrial pain radiating to the genital organs. After examinations, a splenic lesion of liquid density was detected, leading to splenectomy and confirmation of the diagnosis of isolated and diffuse splenic peliosis. Patients with isolated splenic peliosis may be asymptomatic or describe abdominal discomfort and are often diagnosed incidentally during abdominal imaging. This condition is rare and its origin is still unknown.

Keywords: Peliosis, isolated peliosis, ultrasound, Computed tomography, rare condition.

INTRODUCTION

Peliosis is characterized by the appearance of multiple cystic cavities filled with blood in the parenchyma of solid organs. This condition of unknown pathogenesis most commonly affects the liver, but also the spleen, lungs, bone marrow, kidneys, and lymph nodes. Isolated splenic involvement is a very unique entity and rare cases have been described in the literature. We report a case of isolated splenic peliosis in a 56-year-old man, confirmed after splenectomy.

CASE REPORT

A 56-year-old man, with no particular medical history, presented with recurrent pain in the left hypochondrium radiating to the genital organs. Acute renal colic was suspected, with normal blood tests and no associated signs of clinical severity. An unenhanced uro-CT scan was performed to search for a urinary tract stone, which revealed a kidney stone with slight dilation of the pyelocalyceal cavities. However, a liquid-density lesion of the spleen was also noted (Figure 1), which prompted the administration of contrast agent to better characterize the lesion.

After the injection of the contrast product following a three-step protocol: a time without injection, an arterial time at 15s and portal time at 60s. The scanner objectified a subcapsular splenic lesion, with lobulated contours, measuring 58x54mm, seat of multiple partitions of variable size, the thickest of which measuring 4mm in maximum thickness and some of which are enhanced after injection of the contrast product (Figure 2 & 3). The liver was normal in size and appearance with no nodular lesions or pathological enhancement (Figure 3). The echographic complement showed a spleen of normal size, with regular outlines, seat of a cystic formation, lobulated outlines, with anechoic content, seat of multiple thin and thick septa, some of which are vascularized on color Doppler. The liver was of normal size, regular contours, homogeneous with no associated nodular lesion.

The diagnosis that was considered first was that of a splenic cystic lymphangioma. The patient underwent an ultrasound after six months, which showed a slight increase in size of the isolated splenic lesion, prompting a splenectomy.

The pathological examination received a surgical specimen weighing 700g, in a normal-sized capsule. On sectioning, macroscopic examination revealed a poorly defined, non-encapsulated, multi-cystic lesion measuring 7cm in maximal diameter. The vascular cavities had a thin wall, with a brownish or hematic content. Microscopic examination of this lesion showed vascular cavities, with thin capillary-like walls, anastomosed, and sinusoids. They were lined by a flat and regular endothelium. The pathological examination did not reveal any signs of malignancy in the samples taken. The rest of the splenic parenchyma appeared

normal. A diagnosis of isolated and diffuse splenic peliosis was made.

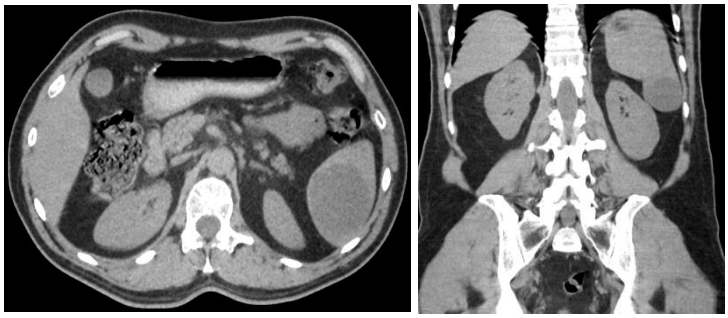

Figure 1: CT scan without injection: axial and coronal section showing an inferior polar splenic lesion with a density close to a liquid density

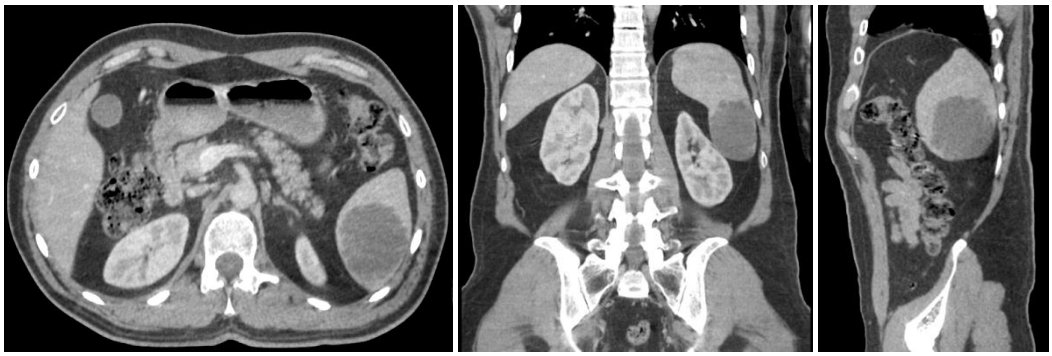

Figure 2: CT with injection (portal time): sub capsular splenic lesion, with lobulated contours, with multiple partitions of variable size, some of which are enhanced after injection of the contrast

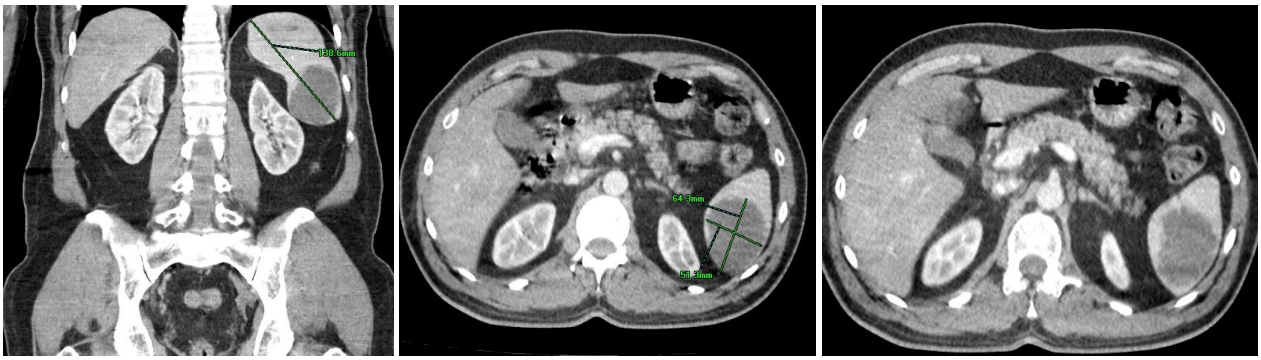

Figure 3: CT with injection (portal time): sub capsular splenic lesion, with lobulated contours, associated with splenomegaly. The liver was of normal size with no detectable nodular lesion

DISCUSSION

The term 'peliosis', the Greek word meaning 'blackish blue', was first used by Wagner in 1861 to describe the gross appearance of lesions found on the cut surface of a liver affected by this ailment [1]. It mainly affects the mononuclear phagocytic system organs. Isolated peliosis of the spleen is extremely rare and occurs in less than 1% of cases at autopsy [2]. It can be idiopathic or associated with infections such as tuberculosis, hematological diseases such as myeloma and myelofibrosis, and the use of androgenic steroids [3].

Patients with isolated splenic peliosis may be asymptomatic or may describe abdominal discomfort. It is often detected incidentally on abdominal imaging or during spontaneous rupture [4]. The latter remains the most serious and potentially fatal complication, due to the occurrence of intra-peritoneal hemorrhage, either spontaneously or following minor trauma [5]. Various radiological examinations help to characterize the lesion despite its non-specific appearance on imaging.

On ultrasound, there are multiple hyperechoic or hyperechoic lesions with poorly delimited edges, of focal localization or sometimes diffuse if the spleen is entirely affected. On the CT scan, low-intensity, spontaneously hypodense foci are observed with enhancement similar to that of hemangiomas, showing centripetal enhancement with late filling [6]. Usually, extracapsular extension is not observed. However, in case of rupture of the richly vascularized cysts, a

subcapsular hematoma and intraperitoneal hemorrhage can be observed on the CT scan [5]. MRI shows lesions exhibit a mixed signal on T2-weighted sequences due to the presence of deoxyhemoglobin and methemoglobin. Sometimes a variable signal can be observed on the T1-weighted sequences reflecting the different stages of the haemorrhage, with no enhancement of the lesions after injection of the contrast product [7].

Rare cases of peliosis have been reported in the literature, most of them consult for pain in the left hypochondrium without other associated signs, such as the case of G.H Chieng *et al*., [1] who took splenic peliosis as the original lesions. Probable metastatic without detectable primary lesions. The other cases arrive in a picture of complications and diffuse pain like the case of Patel *et al*., [8] who diagnosed a hemoperitoneum of great abundance secondary to the rupture of the lesions of the splenic peliosis. Our case is a little unusual because the patient consulted for symptoms of another disease that has no relation to peliosis, and it was only when an incidental finding on a CT scan did we have detected this lesion. The patient did not present with splenomegaly or clinical signs suggestive of splenic rupture.

The diagnosis made on the non-contrast and contrast-enhanced CT scan was a splenic lymphangioma, despite the atypical radiological features. Other organs appeared normal. Due to the lesion's increased size on ultrasound follow-up after six months, and the non-specific nature of the findings on the CT scan, as well as the potential serious complications of invasive procedures like biopsy, surgical management with splenectomy was recommended. The radiological diagnosis of splenic peliosis is a diagnosis of exclusion due to its rarity and non-specific nature. The differential radiological diagnosis of peliosis includes hemangiomatosis, lymphangioma, and angiosarcoma [9].

The diagnosis is generally made macroscopically, and histopathological analysis reveals numerous lakes of different sizes, filled with blood or thrombus, without endothelial lining. Peliosis differs from splenic hemangioma in that the spaces filled with blood are dispersed in a disorderly manner in the red pulp and there is a preferential involvement of the para-follicular zones of the spleen.

CONCLUSION

In conclusion, splenic peliosis is a rare benign tumor that is classified as a vascular neoplasm due to its similar radiological and pathological features. Despite its isolated and rare nature, clinicians and radiologists should always be concerned with the possibility of other contributing factors or diseases in order to make an early diagnosis and provide precise management, which can prevent fatal outcomes.

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