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Fever of Unknown Origin and a Splenic Mass: When Infection and Incidental Findings Coalesce

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Introduction

Primary splenic neoplasms are generally categorized into lymphoid neoplasms arising from the white pulp and vascular neoplasms arising from the red pulp. Splenic lymphangiomas are rare, benign, slow-growing neoplasms arising from vascular elements. These tumors are usually encountered during childhood as an abdominal mass. In adults they are often noted as an incidental, asymptomatic finding, detected by abdominal ultrasonography or by an abdominal computed tomography (CT) scan [1]. When symptomatic, the clinical features typically include abdominal pain, nausea, and abdominal distention [2]. The current case presentation describes a case of incidentally detected splenic lymphangioma in a 56-year-old male presenting with constitutional symptoms including low-grade fever and night sweat. The patient concurrently had abdominal pain and a splenic mass identified by ultrasonography done prior to hospitalization. The constellation of B-symptoms along with a splenic mass necessitated ruling out the differential diagnosis of a lymphoproliferative disease. Accordingly, the patient underwent double contrast abdominal CT, which revealed the characteristic findings of a splenic lymphangioma. The diagnosis was ultimately based on radiography.

Patient Description

A 56-year year-old man was admitted to internal medicine due to low grade fever and dull epigastric pain, beginning 10 days prior to admission, accompanied by night sweats and loss of appetite. An abdominal ultrasonography performed prior to admission, at an outpatient clinic, revealed an enlarged spleen measuring 14.2 cm. A multi-locular mass measuring 6.6 cm was identified within the spleen, composed of cystic areas bordered by gross septations. Aside from abdominal cramps, the patient reported no other symptoms potentially related to the splenic mass. The patient's past and family medical history was unremarkable. Physical examination revealed no abdominal tenderness upon palpation. No abdominal mass, splenomegaly or lymphadenopathy were identified. The rest of the physical examination was normal. Although the patient's fever did not match the definition of FUO by Petresdorf [3], we decided to initiate a workup that would suit such presentation. Initial laboratory findings included liver function tests that were slightly elevated: SGOT – 57 IU/L, SGPT – 68 IU/L, GGT – 123 IU/L, LDH – 487 IU/L and Alkaline phosphatase – 136 mg/dl. Peripheral blood count, coagulation studies, kidney function tests and C Reactive Protein [CRP] levels were all within the normal range. During hospitalization, CMV-specific IgM titers were increased. An abdominal Computed Tomography CT (Figures 1A and 1B) with both oral and intravenous contrast material revealed a multi-locular cystic lesion in the spleen, located in the sub-capsular region, with sharp, well-defined borders and thin septations. Small foci of calcifications were noted in the septations and in the wall of the lesion. The septations within the lesion were slightly enhanced after contrast material administration, but no solid contrast-enhancing component was identified. According to the clinical and radiological findings, a diagnosis of infectious mononucleosis in a patient with an asymptomatic splenic lymphangioma was done.

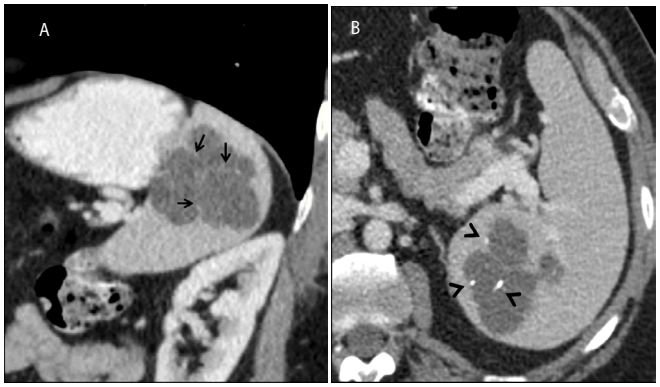

Figure 1: (A): Axial CT image after administration of intravenous contrast material

(B): Sagittal reconstruction demonstrate a sub-capsular cystic lesion with thin septations (black arrow) and mural calcifications (arrow head)

The patient was instructed to rest and avoid contact sports for 6 weeks duration. Follow up CT was scheduled in 6 weeks.

Discussion

Lymphangioma is a benign congenital neoplasm caused by a malformation of the lymphatic system. The incidence of lymphangioma occurring after 20 years of age is very low, commonly occurring before the age of 2 years. The most commonly involved sites are the cervix (75%) and axilla (20%). Infrequently encountered sites are the mediastinum, adrenal gland, kidney, bone, omentum, gastrointestinal tract, retro peritoneum, spleen, liver and pancreas. There are two suggested pathophysiological mechanisms potentially accountable: a). abnormal congenital development, and b). bleeding or inflammation within the lymphatic system which causes an obstruction of lymph vessels; both potentially leading to limited communication with the lymphatic system and thus generation of lymphangiomas [4]. The pathologic appearance ranges from single and multiple nodules to diffuse lymphangiomatosis. The tumors are generally divided to three types – capillary, cavernous and cystic, depending on the size of dilated

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lymph vessels. Lymphangiomas of the spleen are rare and can belong to any of these three subtypes. Splenic cystic lymphangioma can present as a single cystic cavity, but more often consists of multiple cysts of variable sizes separated by internal septations. The cysts are composed of abnormally dilated lymphatic vessels lined with a single layer of endothelium which contain proteinaceous fluid (lymph). The septations are composed of fibrous connective tissue with sparse vessels, which may calcify.

Lymphangiomas usually present as an incidental finding in patients undergoing routine radiologic examination. Clinical manifestations of the tumors include left upper quadrant pain, abdominal distension, loss of appetite, weight loss, nausea, vomiting and a palpable mass. The severity of the symptoms, primarily left upper quadrant abdominal pain, is correlated with the size of the lesion. The lesion can also present with a variety of complications, including portal hypertension, hemorrhage, coagulopathy and hypersplenism. Physical examination can be unremarkable or reveal a palpable mass in the left upper quadrant. Routine laboratory tests, chest and abdominal radiographs are usually unremarkable. On ultrasonography the cysts are anechoic or hypo-echoic. The septa are hyper-echoic and vasculature may be apparent at color Doppler. On CT scan these lesions range from a few millimeters to centimeters in diameter, and are typically sub-capsular. The typical appearance is that of a mass composed of thin-walled, well-marginated cysts with low attenuation that may contain mural calcifications. After injection of contrast material the cystic fluid does not enhance, which may hinder detection. Septations often exhibit moderate enhancement. Magnetic Resonance Imaging (MRI) typically shows a multi-loculated hyper-intense mass, indicating a cystic lesion on T2-weighted images. The septa are seen as hypo-intense bands compared with the surrounding parenchyma. On T1- weighted images the cysts usually appear hypo-intense. MRI is important for the identification of possible areas of malignant degeneration. Malignancy is suggested by abundance of solid areas within the mass. In such cases, MRI may facilitate the detection of solid areas by offering high contrast resolution and by enhancement after gadolinium injection.

The differential diagnosis is broad and includes lymphoma, infarction, metastases (melanoma, breast, ovarian and lung), true splenic cysts, post-traumatic cysts, echinococcal cysts, and false cysts. True splenic cysts with true epithelial lining usually show no wall enhancement. False splenic cysts are thought to result from splenic hematoma, and may be hard to distinguish from true cysts. Hematomas may present MR signals typical of blood, corresponding to their age. Echinococcal cysts in their early stages demonstrate as a single cyst with regular margins. In late stages they may become multi-loculated with daughter cysts.

The natural history of splenic lymphangiomas is variable. Small lesions are often asymptomatic and incidentally detected. Larger lesions may grow and compress other organs. Splenectomy is recommended for large lesions as the risk of rupture even from minor abdominal trauma is high. The presence of symptoms is another indication for splenectomy [5].

Conclusion

our patient presented with nonspecific constitutional symptoms, and a splenic mass on ultrasound, suggesting a neoplastic disease. A careful clinical approach to the patient, regarding signs, symptoms and further radiological workup revealed the correct diagnosis. This case presents a complex mix of a congenital benign mass with superimposed acute viral disease, confirmed by serology for CMV-specific IgM.

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