

Case Report

Cystic lymphangioma of spleen: an abdominal flabbergast

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ABSTRACT

Lymphangioma is congenital malformation of the lymphatic system. It is usually found in children, occurs mostly in the neck and the mediastinum. It is rarely found in the spleen, mostly asymptomatic or detected incidentally by imaging modalities, however the clinical features of splenic lymphangioma include abdominal pain, nausea, and abdominal distention. In our case report Lymphangioma with cystic component and daughter cysts has radiologists to consider it as other neoplastic diseases of the spleen.

Keywords: Lymphangioma, Benign tumor, Spleen, Splenic neoplasia

INTRODUCTION

Spleen diseases are rare, being splenic abscesses, splenic cysts, benign tumors (hemangioma, lymphangioma and others), and malignant tumors (lymphomas, metastases and others).¹ Primary benign tumors of the spleen are extremely rare and account for less than 0.007% of all tumors.² Splenic lymphangiomata are benign tumors predominantly affecting children, whereas few cases have been reported in adults. Hereby reporting a case of splenic lymphangioma in a 55 years female that presented to our hospital. To our knowledge this is the first case being reported from the Indian subcontinent in over 55 years being unique because of the age of presentation.

CASE REPORT

A 55 years female, known diabetic and hypertensive presented to the surgical department with left upper abdomen pain of one month duration, of dull aching nature associated with nausea. She had no history of vomiting, hematemesis, fever, jaundice or weight loss. She had no previous history of abdomen trauma or any intra-abdominal infection. On examination her vitals were stable. Abdomen examination showed tenderness in

the left hypochondrium and lumbar region. No mass or organomegaly was palpable. Rest of the systemic examination was normal. Peripheral blood count, coagulation studies, liver and kidney function tests were all within normal limits. Ultrasound abdomen showed enlarged spleen with well defined, multiseptated cystic lesion of about 8 x 7 cm. CECT abdomen showed enlarged spleen with well-defined cystic lesion and multiple peripheral daughter cysts over the upper pole, 7 x 6.8 x 6 cm, suggesting hydatid cyst of spleen. With the clinical and radiological diagnosis of hydatid cyst of spleen, patient was stabilized and taken for elective splenectomy.

Intra-operative findings were enlarged spleen measuring 12 x 9 cm with cystic lesion of size 6*4 cm occupying the hilum and upper pole of spleen. Rest of the spleen was normal. Other solid organs, hollow viscous and peritoneum were found to be normal. Her postoperative period was uneventful. HPE report showed splenic parenchyma with thickened capsule, and lymphoid stroma with multiple cystic dilated spaces, lined by attenuated endothelium, enclosing proteinaceous fluid admixed with lymphocytes suggestive of cystic lymphangioma of spleen. After one year of outpatient

follow-up, she continues to be asymptomatic and is in good health.



Figure 1: CECT abdomen showing cystic lesion of spleen.



Figure 2: Specimen of spleen with cystic lesion.



Figure 3: Cut specimen showing cystic lesions with peripheral cyst.

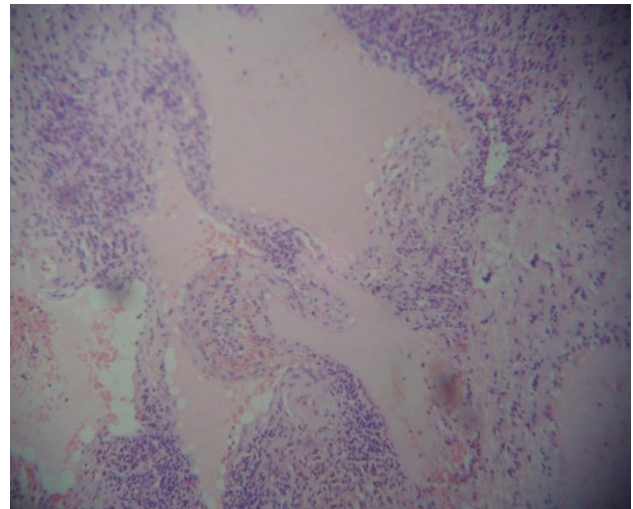


Figure 4: HPE showing lymphoid stroma with cystic dilations with lymphocytes.

DISCUSSION

Splenic Lymphangiomas (SL) are benign cystic tumors resulting from congenital malformations of the lymphatic system that appear as single or multiple lesions of the spleen.³⁻⁵ Lymphangioma were first described by Rodenber in 1828.³ Histologically, lymphangiomas are classified into three subtypes according to the congenital dilated lymphatic channels: capillary (super-microcystic), cavernous (microcystic), or cystic (macrocytic).SL

mainly affects children and rarely manifest after 20 years of age.⁶

The clinical features of SL are abdominal pain, vomiting and a palpable mass, although these tumors are asymptomatic in most patients.^{2,7,8} In a series of seven cases, Komatsuda et al.³ showed a close relationship between the occurrence of symptoms and the size of the spleen. However, cases presenting with no symptoms have also been reported.^{7,9} Physical examination may be normal or reveal a palpable mass in the left upper quadrant. Routine laboratory tests and simple chest and abdominal X-rays generally shows no abnormalities.⁶ The differential diagnosis is extensive and includes lymphoma (a more common malignant tumor), infarction, septic embolism, metastases (melanoma, breast, ovarian and lung cancer), and splenic cysts.¹⁰ US usually shows hypoechoic spaces that contain internal echoes. Low-density, well-delimited subcapsular cysts with thin walls that may contain mural calcifications are generally visualized by CT, suggesting a diagnosis of cystic lymphangioma. A “Swiss cheese” appearance of the spleen has been considered to be pathognomonic.⁶

The treatment of choice for intra-abdominal lymphangiomas, including SL, is complete surgical resection.⁴ However, surgical resection is recommended immediately after establishment of the diagnosis since a growing incidence of complications is observed over time, such as infection, hemorrhage, intestinal obstruction and tumor growth.⁴ The prognosis of intra-abdominal lymphangioma after resection is favorable. Recurrence is the main complication, which is demonstrated in 9.5% of patients, frequently after incomplete resection.⁴

CONCLUSION

In conclusion, dynamic contrast-enhanced CT is a useful method for evaluating splenic lesions. Correlation of radiologic features with clinical and histologic findings is needed to confirm the diagnosis. It is important to understand and recognize the spectrum of pathologic and imaging features of cystic splenic lesions.

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