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Case Report

Splenic cystic lymphangioma in adults: A rare case report from Syria

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ABSTRACT

Introduction and importance: Splenic lymphangiomas are an extremely rare entity that is mainly diagnosed in children. They are often found in the neck and axilla region. Cystic lymphangioma in the abdomen is unusual and the spleen is an exceptional location for lymphangiomas.

Case presentation: We report a case of a 73 - year - old woman who presented with abdominal pain in the left upper quadrant for a month. Abdominal imaging studies show multiple splenic cysts. Splenectomy was performed and the specimen was sent for histopathologic examination. Microscopic inspection revealed splenic lymphangioma.

Clinical discussion: Splenic lymphangiomas are uncommon lesions. Most cases have been reported in children. The head, neck, and axillary regions are the most common sites, whereas lymphangioma in the spleen is exceptional.

Conclusion: Splenic lymphangiomas are uncommon benign tumors that are rarely seen during adulthood. Splenectomy is the preferred treatment.

1. Introduction

Lymphangiomas are rare benign tumors of the lymphatic vessels, found mainly in children but rarely in adults [1]. The most common sites of these neoplasms are the neck (75%) and axilla (20%) [1,2]. The Spleen is an exceptional location for lymphangiomas [2,3]. The clinical picture is variable; wherein larger lesions can result in splenomegaly with left upper quadrant pain, while small lesions are often incidentally detected through imaging studies [4]. In this report, we describe a case of splenic cystic lymphangioma in an adult woman.

This case report has been reported in line with the SCARE criteria 2020 [5].

2. Case presentation

A 73 - year - old woman, with a history of hypertension for 30 years on bisoprolol, presented to the emergency department at Tishreen University Hospital in 2022 with abdominal colic pain in the left upper quadrant for a month. Her pain did not respond to analgesics and there was no comfortable position. She did not have a fever, weight loss, nausea, or vomiting. The patient was a nonsmoker and did not consume alcohol. She had no history of allergies. On clinical examination, a tender mass in the upper left quadrant of the abdomen was palpated. Routine blood tests with normal limits. Abdominal Ultrasound showed multiple splenic cysts [Fig. 1A]. The liver, biliary tree, and pancreas appeared normal. Abdominal CT revealed moderate splenomegaly with multiple thin-walled, low-density subcapsular cysts. Some of the cysts had calcified walls and the largest one measured 4.2 cm [Fig. 1B]. Diagnosis of hydatid cysts was suspected and an echinococcus granulosus antibodies test was done, but the result was negative. Therefore, the patient underwent a splenectomy. The specimen was sent to the pathology department. The excised spleen measured 19 cm × 11 cm × 7 cm and weighed 590.5 g. The outer surface was irregular and brownish grey colored with multiple nodular formations. Gross sections showed cystic cavities of different sizes. The largest one measured was 4.5 with some wall calcifications [Fig. 2]. The rest of the splenic parenchyma had

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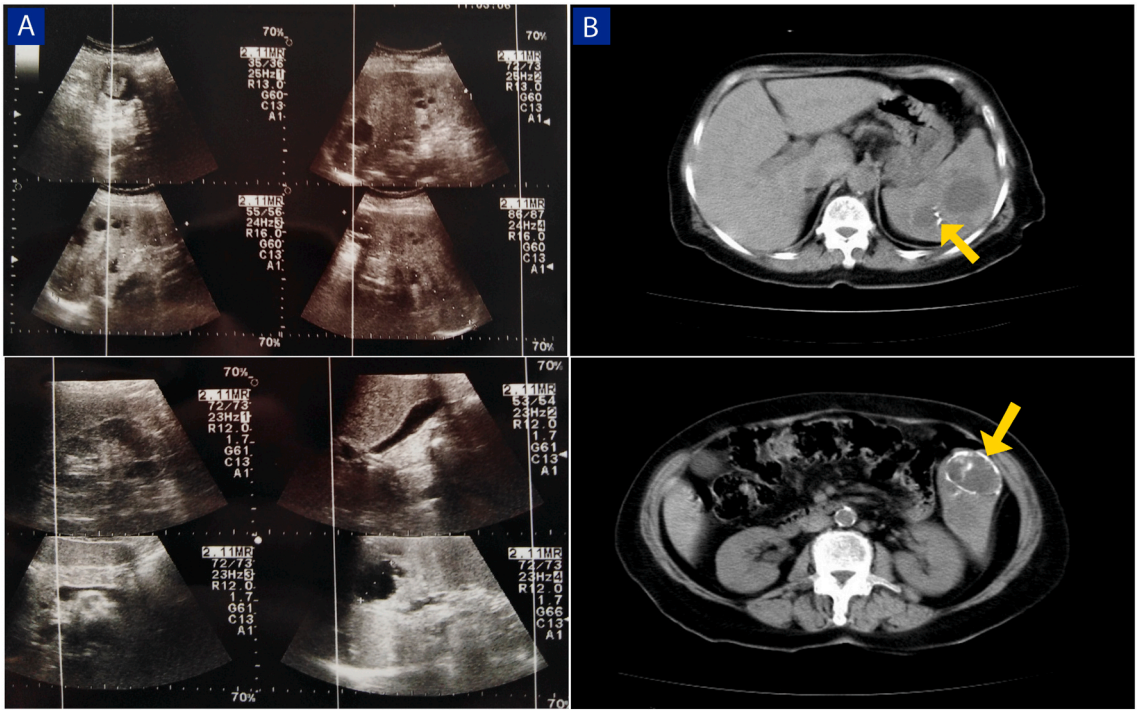

Fig. 1. (A) Ultrasound shows multiple splenic cysts (B) CT scan image demonstrates multiple thin-walled, low-density subcapsular cysts in the spleen. The walls of some cysts were calcified (yellow arrow). . (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

Fig. 2. Gross images of the excised spleen are showing (A) Splenomegaly with a brownish-grey colored irregular surface exhibiting multiple nodular formations. (B) Cut sections reveal cystic cavities of different sizes with occasional wall calcifications.

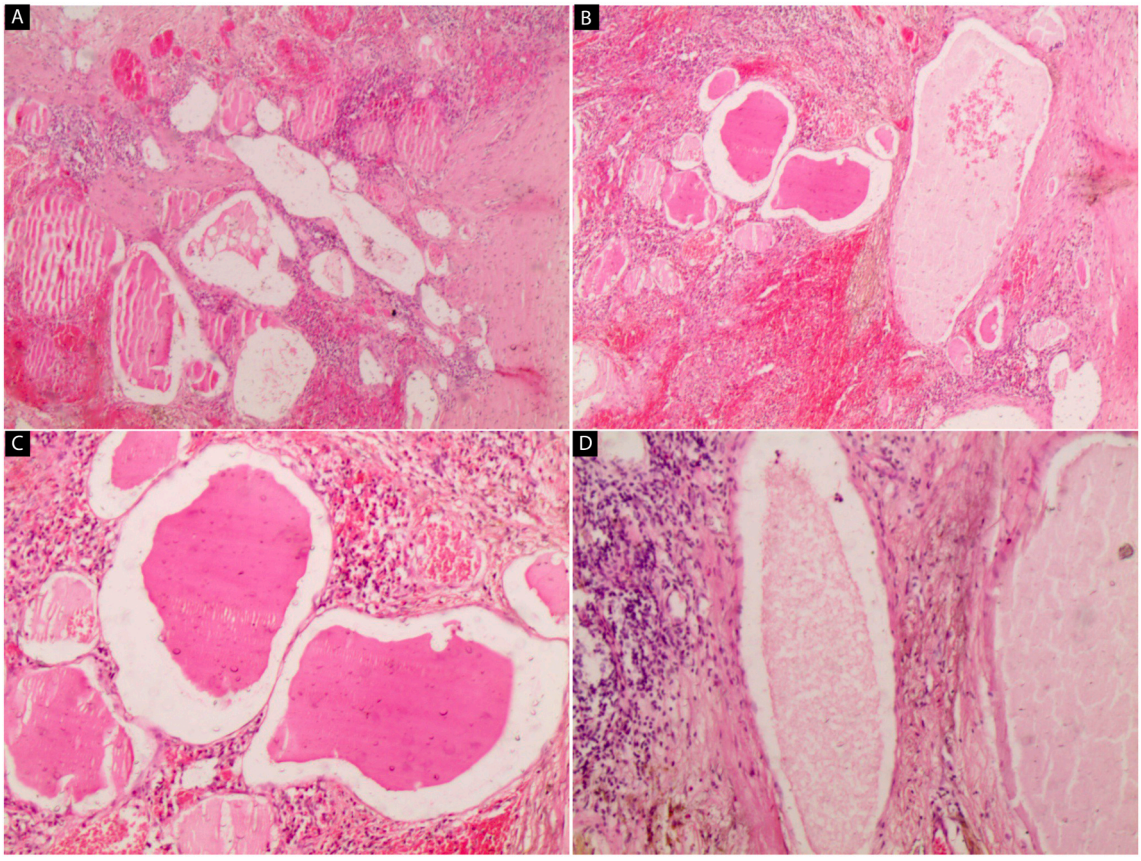

Fig. 3. H&E stain (A–D): Microscopic images of the spleen. (A and B) Multiple vascular spaces are filled with proteinaceous fluid (x 40). (C and D) The spaces are lined by flattened to cuboidal endothelial cells (x 100).

no remarkable changes. Microscopic examination revealed vascular spaces lined by flattened to cuboidal endothelial cells. The lumens were filled with proteinaceous fluid. [Fig. 3]. The patient had a stable recovery and she was discharged few days after the surgery.

3. Discussion

Splenic lymphangiomas are defined as developmental malformations of the lymphatic system [6,7]. Most cases have been reported in children and are generally diagnosed before the age of 2 years. Splenic lymphangiomas in adults are very rare [3,6]. Most lesions are seen in the neck and axillary regions, whereas splenic lymphangioma is extremely rare [3,6]. Clinically, it is asymptomatic in the majority of cases. On the other hand, these lesions may produce significant splenomegaly which can be complicated by hemorrhage, consumptive coagulopathy, hypersplenism, and even portal hypertension [8]. Imaging studies including abdominal ultrasound, CT scan, and MRI are useful tests in the diagnosis of splenic cysts [4,7]. Ultrasound shows a hypoechoic cystic lesion with multiple septa and calcifications. The CT scan reveals more details on the size and nature of the lesion [3,4]. Magnetic resonance has the same advantages as computed tomography. The differential diagnoses of splenic lymphangioma include hydatid cysts, chronic infection, hemangioma, lymphoma, metastasis, and pseudocysts of the spleen. Parasitic infection with echinococcus granulosus is the main cause of cystic lesions of the spleen [9]. In our case, the first clinical and radiological diagnosis was hydatid cysts, but it was ruled out after performing laboratory tests and pathological examination. However, splenic cystic lymphangioma tends to be located in the subcapsular region and it has a thick fibrous wall with internal fibrous trabeculae [6,10]. Microscopically, cystic lymphangioma consists of a single layer of flattened endothelium-lined spaces filled with eosinophilic proteinaceous material. Hyalinization and calcification of the fibrous connective tissue may be present [11]. The treatment of choice for splenic lymphangiomas is total splenectomy, particularly in case of large lesions, in order to avoid potential complications such as splenic rupture, hemorrhage, or hypersplenism, and to exclude the possibility of malignant lesions [1–3]. Other therapeutic methods, including aspiration, and drainage, showed insignificant results [6,8,12].

4. Conclusion

In elderly patients, cystic lymphangioma, in spite of its rarity, should be one of the differential diagnoses of cystic lesions of the spleen. Ultrasound and CT scan are useful in diagnosis. However, the pathological examination of the excised specimen is essential for an accurate diagnosis.

Provenance and peer review

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Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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Ethical approval

No ethical approval was needed for this case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Author contribution

Moatasem Hussein Al-janabi: study design, data collections, data analysis, and writing. Riham Abodest: study design, data analysis, and writing. Majd Adnan Alshabab: data analysis, and writing. Samir Kanaan: data collections. Rana Issa: in reviewing the manuscript.

Registration of research studies

Not applicable.

Guarantor

Rana Issa.

Declaration of competing interest

The authors have no conflicts of interest to declare.

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