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Splenic Cystic Lymphangioma in Adults: Diagnostic Challenges and Surgical Management: Report of Two Cases

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Abstract **Case Report**

Lymphangiomas are benign tumours that are considered to be congenital malformations of the lymphatic system and most commonly affect children, with only a few cases seen in adults. These lesions are typically found in the neck and axillary region, but are rarely found in the spleen. Here we report two unique cases of splenic cystic lymphangioma. The first case was a 46-year-old man with epigastric pain and discomfort in the left hypochondrium associated with a weight loss of 10 kg, and the second was a 22-year-old woman with non-specific, low intensity but persistent pain and a multilocular cystic lesion identified on imaging. These cases highlight the importance of considering splenic lymphangioma in the differential diagnosis of cystic splenic lesions and the importance of accurate diagnosis by imaging and histology, and demonstrate the effectiveness of surgical intervention.

Keywords: Splenic lymphangioma, Cystic lesions, Adult cases, Diagnosis, Surgical intervention.

INTRODUCTION

Splenic lymphangioma is a very rare disease, predominantly diagnosed in children, with adult involvement being the exception [1]. They are typically located in the neck and axillary regions. Abdominal cystic lymphangiomas are rare, making the spleen an exceptional site for these lesions. Splenic lymphangiomas are rare benign tumors characterized by cystic dilatation of lymphatic vessels within the splenic parenchyma. Therefore, we report two cases of splenic cystic lymphangioma.

CASES REPORTS

Observation N°1

Mr. A.H. is a 46-year-old patient. He underwent surgery for peritonitis due to a perforated ulcer at the age of 41. His clinical history began a few months earlier with epigastric pain and discomfort in the left hypochondrium, accompanied by a weight loss of 10 kg.

Clinical examination was unremarkable except for the weight loss. Esophagogastroscopy was normal. Abdominal ultrasound showed splenomegaly with a heterogeneous cystic mass without deep lymphadenopathy. Abdominal CT scan showed a 5 cm splenic lesion with cystic appearance and peripheral enhancement after contrast injection, associated with abnormal thickening of the gastric wall (Figure 1). Hydatid serology was negative and complete blood count was unremarkable.

The clinical context and scan data raised suspicion of malignancy, such as lymphoma or splenic metastasis. The patient underwent a repeat midline laparotomy. Abdominal exploration revealed homogeneous splenomegaly. The gastric wall appeared normal and there was no evidence of abdominal tumor syndrome. The patient underwent splenectomy (Figure 2), and the diagnosis of splenic lymphangioma was made postoperatively based on histologic examination of the surgical specimen.

The course was marked by the occurrence of a midline hernia six months later, which was treated with surgical prosthetic repair.

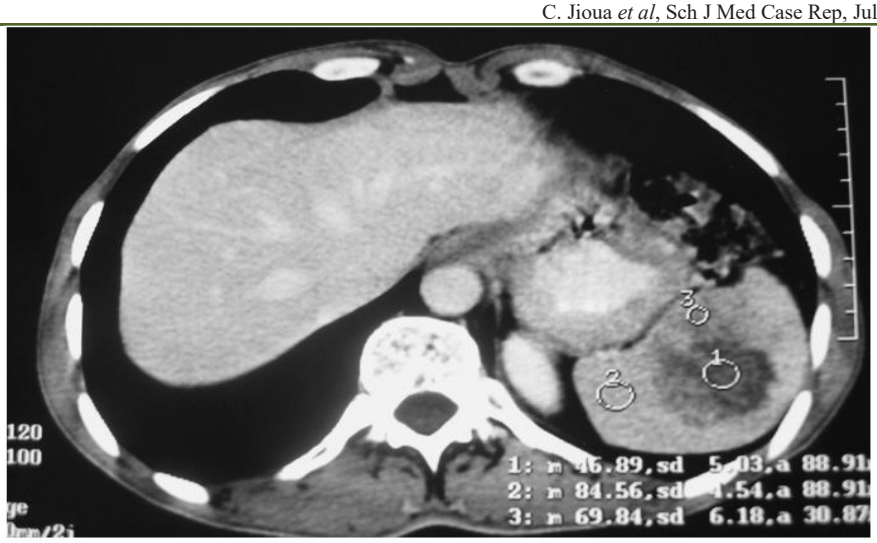

Figure 1: Abdominal CT scan showing a cystic splenic lesion, with peripheral enhancement after contrast injection, associated with abnormal thickening of the gastric wall

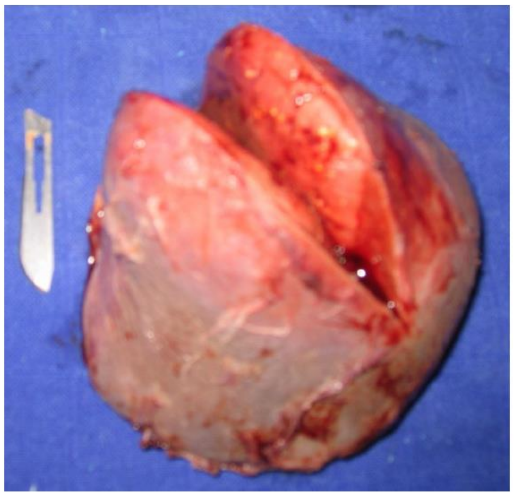

Figure2: Surgical specimen showing the spleen and a section of the lymphangioma

Observation N°2:

Ms. N.B. is a 22-year-old patient with no significant past medical history. Her clinical history began 2 months ago with nonspecific, low intensity but persistent pain, sometimes diminishing to mild discomfort in the left hypochondrium, without digestive symptoms and without affecting her general condition.

The clinical examination on admission was completely normal. The pain prompted an abdominal ultrasound, which revealed a well-defined multilocular cystic lesion in the spleen measuring approximately 6 cm.

A follow-up CT scan confirmed the presence of the splenic lesion. It also showed a second retroperitoneal cystic lesion measuring 4 cm near the lower pole of the left kidney and a third cystic lesion measuring 2 cm on the left ovary. Hydatid serology was negative and tumor markers (CA19-9, ACE, CA125) were normal.

The patient underwent laparoscopic surgery. She was positioned in strict right lateral decubitus for a lumbar approach after orotracheal intubation and bladder catheterization. A Veress needle was used to create pneumoperitoneum and trocars were inserted under visualization. Four trocars were used for splenectomy, placed along the left costal margin. Splenectomy was performed using an anteroposterior approach with a dissecting hook and Ligasure. The anatomical specimen was left in the HCG.

To address the retroperitoneal lesion, the patient was maintained in the same position with a slight retrograde tilt. The first step was to complete the dissection and lower the left colic angle and descending colon, and identify the gonadal vessels and left ureter. The lesion was approximately 50 mm in diameter with a whitish chylous content and a very thin, fragile wall. Careful dissection with the dissecting hook allowed its complete removal, resulting in a small rupture of its capsule.

The left ovary was approached with the patient in the same position in light Trendelenburg and using the same trocars. The ovarian cyst was simple, with a thin, homogeneous wall without septa, measuring approximately 2 cm. Its complete removal was achieved. The retroperitoneal and ovarian lesions were placed in an endobag prior to extraction to avoid loss in the peritoneal cavity due to their size. The spleen was extracted intact through a small Pfannenstiel incision after placement of a Redon drain in the HCG.

Postoperative recovery was uncomplicated and the patient was discharged from the hospital on postoperative day 3.

About the pathological examination

- Spleen: splenectomy specimen measuring 12x6x2 cm, with cystic appearance and hemorrhagic changes; a whitish lesion measuring 1 cm was noted, located 1 mm from the capsule.
- Left pararenal cyst: presence of several fragments measuring from 0.4 to 1.8 cm.
- Pelvic cyst: a fragment measuring 3x2x2 cm, with a cystic appearance and a very thin capsule.

The morphologic appearance is identical in the three sampled sites, corresponding to cystic lymphangiomas, without histologic signs of malignancy.

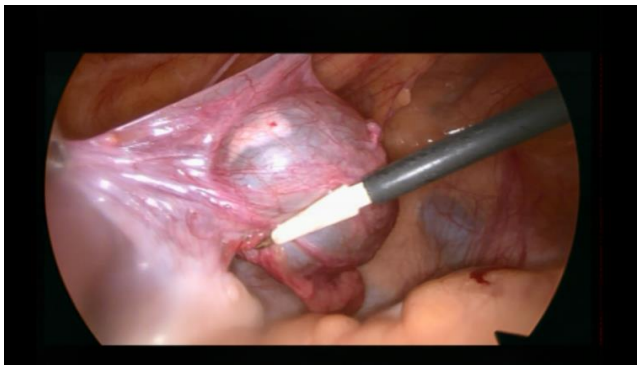

Figure 3: Laparoscopic operative view showing the pelvic cystic lesion before its resection

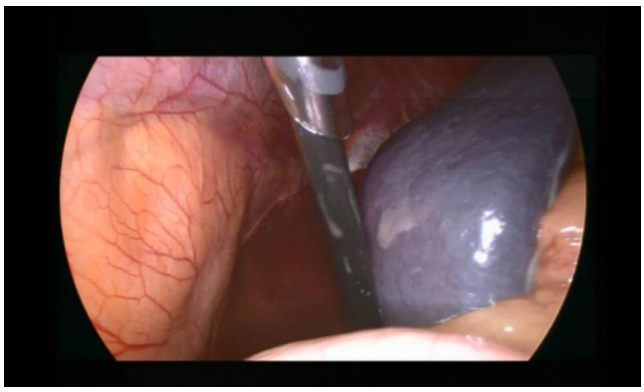

figure 4: Operative view showing the spleen left in the HCG after its removal, prior to treatment of the retroperitoneal and pelvic lesions

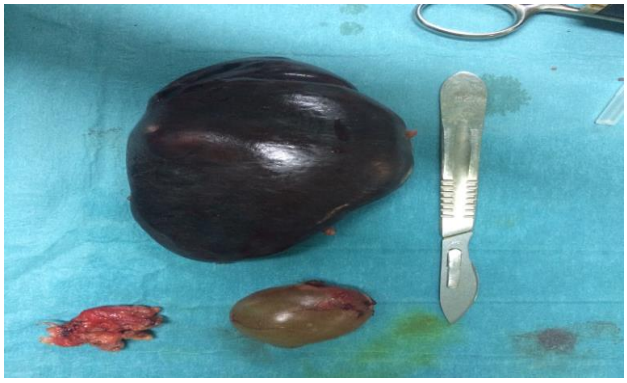

Figure 5: Surgical specimens of the spleen, retroperitoneal cyst wall and pelvic cyst, and the pelvic cyst

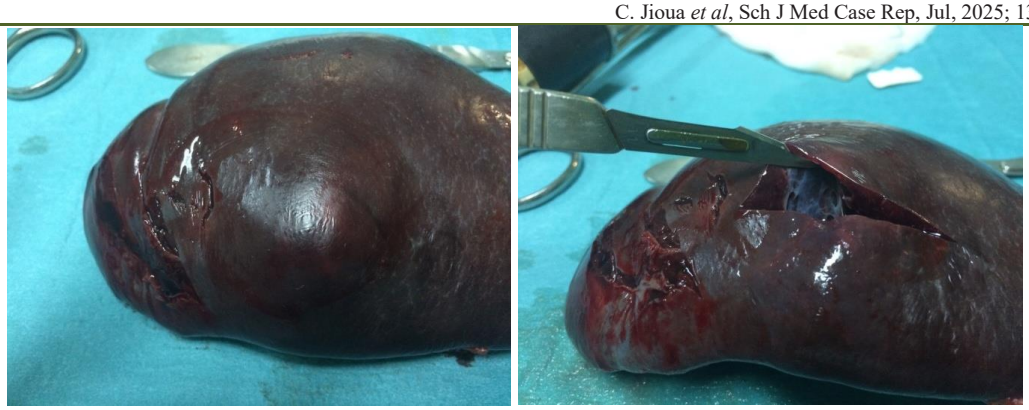

Figure 6: surgical specimens of the spleen

DISCUSSION

Lymphangiomas are benign tumors resulting from a congenital malformation of the lymphatic system that results in lymphangiectasis due to the lack of normal communication between the lymphatic ducts. These ducts terminate at the base of a sac and gradually expand to form a cyst (2-3). They are usually located near the lymph node areas, namely the cervical and axillary regions. In the abdomen, lesions preferentially affect the mesentery and omentum, with retroperitoneal localization being the second most common site.

It may also involve the liver, pancreas, kidney, adrenal gland, colon or duodenum, with splenic localization being rare. Approximately 90 cases have been reported in the literature [4]. Our study clearly demonstrates the rarity of this localization, as we observed only two cases in a period from 2014 to 2024. It is a slowly growing tumor typically observed in childhood and rarely detected after the age of 20 years [5].

Clinically, most isolated splenic lymphangiomas are asymptomatic and are discovered incidentally during an abdominal ultrasound or CT scan performed for other reasons. In large lesions, symptoms may include left upper abdominal pain, nausea, vomiting, anorexia, abdominal distention, and possibly palpable splenomegaly. However, these lesions can lead to significant splenomegaly, which may be associated with complications such as bleeding, consumptive coagulopathy, hypersplenism, and even portal hypertension [6].

Imaging studies such as abdominal ultrasound, CT, and MRI are valuable in the diagnosis of splenic cysts. [7-8] Ultrasound shows a hypoechoic cystic lesion with multiple septa and calcifications, while CT provides additional information about the size and characteristics of the lesion. [9-10] Magnetic resonance imaging provides the same benefits as computed tomography.

Fine-needle aspiration biopsy is contraindicated because of the risk of bleeding and the limited tissue sample available for accurate diagnosis. [11] The diagnosis is confirmed by histopathology. The diagnosis is confirmed by histopathologic examination after spleen removal. [12] Histologic analysis of capillary, cavernous, and cystic lymphangiomas shows a single layer of flattened epithelium lining spaces between fat, fibrous, and lymphatic structures filled with eosinophilic proteinaceous material. Hyalinization and calcification of the fibrous connective tissue may also be present.

The differential diagnosis of splenic lymphangioma often includes other solid and cystic lesions of the spleen, such as hemangiomas, mesothelial cysts, and parasitic cysts (most commonly due to Echinococcus granulosus), lymphomas, and metastases. Total splenectomy is the treatment of choice for splenic lymphangiomas, especially for large lesions. Partial splenectomy is becoming more common, but is associated with an increased risk of recurrence in the remaining spleen.

Both open and laparoscopic techniques can be used, depending on factors such as the patient's characteristics, the surgeon's experience, and the extent of the splenomegaly. The laparoscopic approach is generally preferred for spleens with normal to moderate enlargement, while the open method is typically chosen for significantly enlarged spleens. The prognosis for splenic lymphangioma after complete resection is favorable, with a very low risk of malignant transformation, and postoperative surveillance typically focuses on complications related to splenectomy. [13]

CONCLUSION

Splenic tumors are rare, and splenic lymphangiomas occurring in adulthood are particularly uncommon. Preoperative diagnosis can be challenging in asymptomatic patients. Ultrasound and CT scans are helpful in diagnosis, but pathologic examination of the excised specimen is essential for accurate diagnosis. Once diagnosed, surgery is often the most effective treatment option, and laparoscopic splenectomy can help minimize potential complications.

REFERENCES

1. Solomou EG, Patriarheas GV, Mpadra FA, *et al* (2003) Asymptomatic adult cystic lymphangioma of the spleen: case report and review of the literature. Magn Reson Imaging 21:81–4
2. 2. Al-Shaikh S.A., Mubarak A.M., Harb Z.F. Splenic lymphangioma in an adult. Saudi Med. J. 2017;38(11):1148–1152. doi: 10.15537/smj.2017.11.20625.
3. Yang F., Chen W.X. Splenic lymphangioma that manifested as a solid-cystic mass: a case report. World J. Gastroenterol. 2013;5:781–783. doi: 10.3748/wjg.v19.i5.781.
4. I. Korbi, Faouzi Noomen, S. Mosbahi, O. Toumi, Mohamed Hammami, Ammar Mahmoudi, Mohamed Nasr, Wassim Kallel , Khadija Zouari , Hamdi A: LYMPHANGIOME KYSTIQUE DE LA RATE À PROPOS D'UNE OBSERVATION . Le journal de coelio-chirurgie. No 81. Mars 2012.
5. Kaza R., Zara S., Al-Hawary M.M., Francis I.R.: Primary and secondary neoplasms of the spleen. Cancer Imaging 2010; 10: pp. 173-182.
6. 6.Kim S.Y., Kwon H.J., Park H.W., Lee S.Y., Son B.H., Kim M.S. Multiple cystic lymphangiomas of the spleen: radiologic and histopathologic findings. J. Med. Ultrason. 2015;42:409–412. doi: 10.1007/s10396-015-0612-x.
7. Rodrı´guez-Montes JA, Collantes-Bellido E, Marı´n-Serrano E, Prieto-Nieto I, Pe´rez-Robledo JP. Splenic lympangioma. A rare tumour. Presentation of 3 cases and a literature review. Cir Cir. 2015;84:154–9 [in Spanish].
8. Beltran MA, Barrı´a C, Pujado B, Oliva J, Contreras MA, Wilson CS, *et al*. Gigantic cystic splenic lymphangioma. Report of one case. Rev Med Chil. 2009;137:1597–601.
9. Perez A., Perez M., Yuga A.C., Viray B. Splenic lymphangioma in adulthood: a case report. Int. J. Surg. Case Rep. 2020;67:250–253. doi: 10.1016/j.ijscr.2020.01.061.
10. Ousmane T., Mamadou F.P., Sitor S.I., Abdou N., Madieng D. Splenic lymphangioma,International. J. Surg. Case Rep. 2019;62:40–42. doi: 10.1016/j.ijscr.2019.07.078. ISSN 2210-2612.
11. .Alkofer B, Lepennec V, Chiche L. Splenic cysts and tumours: diagnosis and management. Jchir (Paris) 2005;142:6–13. 10.1016/S0021-7697(05)80830-0
12. 12.Anadol AZ, Oguz M, Bayramoglu N *et al*. Cystic lymphangioma of the spleen mimicking hydatid disease. J Clin Gastroenterol 1998;26:309–11. 10.1097/00004836-199806000-00021
13. 13.George Verghese B, Kalvehalli Kashinath S, Kanth RR . Lymphangioma of the spleen—a rare tumor rarely seen in an adult: a case report and a comprehensive literature review.Euroasian J Hepatogastroenterol 2013;3:64–9.