



Laparoscopic splenectomy for a large multilocular splenic cyst with elevated CA19-9: Report of a case

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ABSTRACT

INTRODUCTION: Because splenic cysts are rare, a definitive treatment regime for these cysts remains unclear. We report a case of a large multilocular splenic cyst with elevated carbohydrate antigen 19-9 (CA19-9) levels, which was successfully treated with laparoscopic splenectomy.

PRESENTATION OF CASE: A 22-year-old female was admitted to our hospital with severe left upper abdominal pain. Serum CA19-9 level was mildly elevated (65 U/ml). Computed tomography revealed a 25-cm long spleen with multilocular cystic lesions, for which an emergency laparoscopic splenectomy was performed. Histological findings revealed that the lesion was a benign true cyst, and immunostaining analyses showed that the epithelium was CA19-9-positive.

DISCUSSION: Although some spleen-preserving approaches have been reportedly used, splenic cyst recurrence usually occurs in true cyst cases, wherein the cyst is incompletely removed. Most reported cases of splenic cysts producing CA19-9 are true cysts.

CONCLUSION: The treatment approach should be decided on the basis of the type, shape, location, and even CA19-9 levels of the splenic cyst.

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1. Introduction

Splenic cysts are rare^{1,2} and classified as either true or pseudo cysts depending on the presence or absence of a cellular lining in the cystic wall. Carbohydrate antigen 19-9 (CA19-9) is a well-known tumor marker of pancreatic adenocarcinomas; however, even benign diseases often exhibit elevated serum CA19-9 levels. Approximately 30 cases of CA19-9-positive splenic cysts have been reported in the literature,³ and almost all cysts with elevated CA19-9 levels were true cysts.

Because of the rarity of splenic cysts, a definitive treatment regime for these cysts remains unclear. We report a case of a large, multilocular splenic cyst with elevated CA19-9 levels, successfully treated with laparoscopic splenectomy.

2. Case report

A 22-year-old female was admitted to our hospital with severe left upper abdominal pain. She had no significant past medical, surgical, or traumatic history. Physical examination revealed severe tenderness and a palpable fist-sized mass in her left upper abdomen. Hematological examination was normal, and serum tumor marker levels of CA19-9 were elevated to 65 U/ml

(normal: 0–37 U/ml). Ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI) revealed splenomegaly (a 25-cm long spleen) with multilocular cystic lesions (Fig. 1a and b). CT findings also revealed a hematoma in the splenic cyst, but it did not cause the cyst to rupture. However, the patient's symptoms were severe, which required an immediate treatment plan. The splenic cyst was multilocular and the spleen was mostly occupied by the cyst. Therefore a splenic preservation procedure carried the risk of incompletely removal of the splenic cyst, we planned an urgent laparoscopic splenectomy.

Under general anesthesia, the patient was placed in a right semi-lateral decubitus position. On exploration, multilocular cystic lesions of the spleen were adhered to the diaphragm and the left side of the parietal peritoneum, pushing the stomach to the left (Fig. 2). First, the splenic artery and vein were divided using a clipping technique to control bleeding. Next, to reduce the cystic volume, a needle puncture was performed and a brown liquid was drained. The CA19-9 level in the cystic fluid was extremely high (805,570 ng/ml). The spleen was then dissected from the splenocolic ligament using an ultrasonic scalpel and an electric knife. The splenic hilum was dissected with an auto-suturing device. A vinyl bag was introduced into the abdominal cavity to remove the resected spleen; the spleen was fragmented and removed from the abdominal cavity.

Histological findings revealed that the lesion was a benign true-cyst, and immunostaining analyses showed that the epithelium was positive for CA19-9 antibody (Fig. 3).

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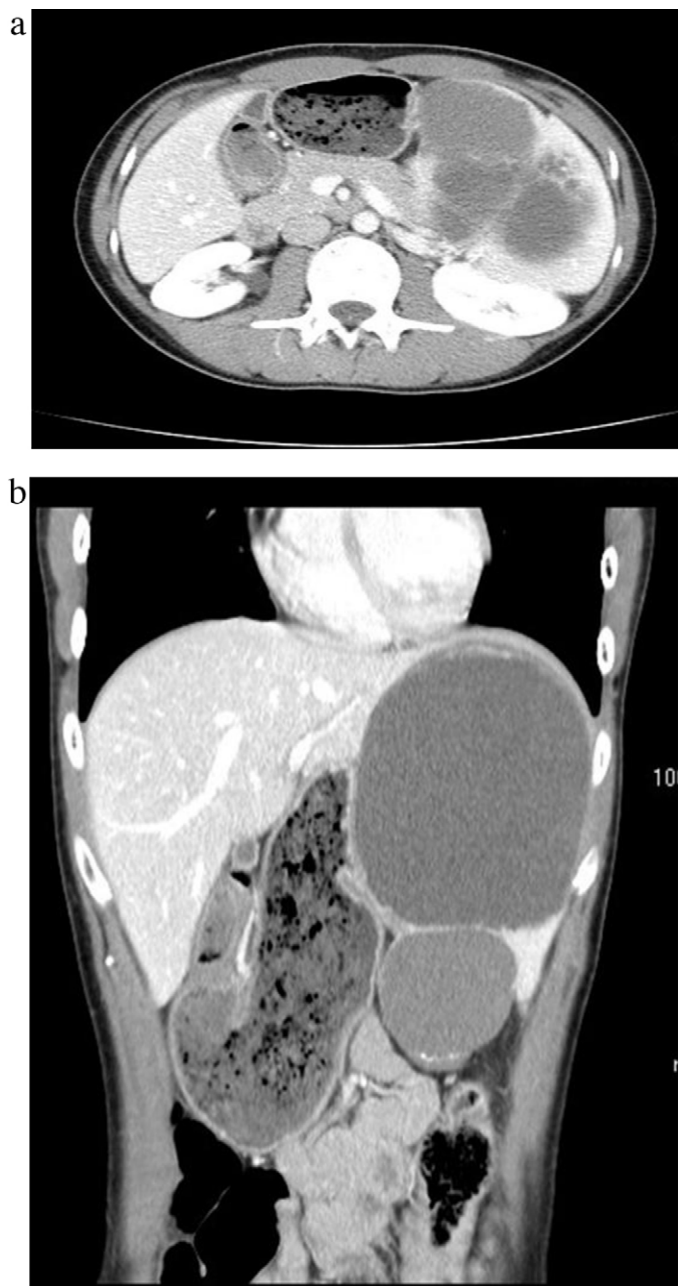


Fig. 1. CT of abdomen shows 25-cm long splenomegaly with multilocular cystic lesion. a is horizontal image, b is coronal image.

The patient was received pneumococcal vaccine and discharged on postoperative day 10 with no complications. On postoperative day 30, the patient’s serum CA19-9 level decreased from 65 to 27 ng/ml.

3. Discussion

Because of the rarity of splenic cysts, a definitive treatment regime remains unclear. Previously, laparoscopic splenectomy was the standard treatment for symptomatic or large splenic cysts.⁴ However, a splenectomy carries a large risk of postoperative infection and thrombocytosis. Thus, spleen-preserving approaches (aspiration, marsupialization, fenestration, decapsulation, cystectomy, partial splenectomy, among others) have been recently proposed. However, these spleen-preserving approaches

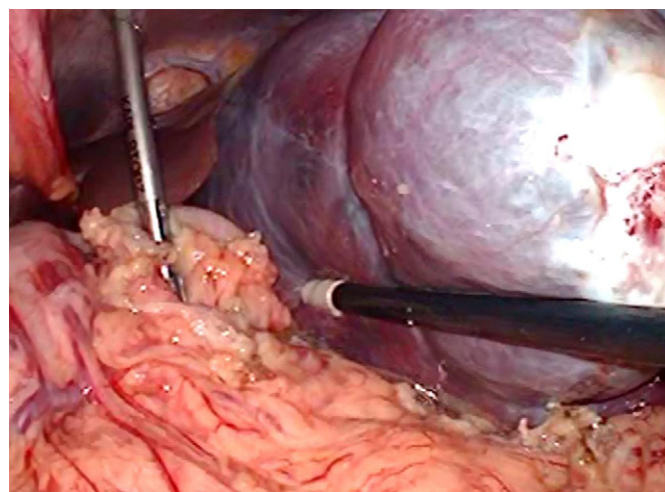


Fig. 2. Laparoscopic view of the splenic cyst.

have some disadvantages. For example, percutaneous cystic aspiration and intracystic injection with tetracyclines or sclerosing substances is associated with high recurrence rates and a risk of abscess formation,⁵ whereas a partial splenectomy is technically difficult.⁶ Splenic cysts are usually observed as benign diseases in young adults.^{7,8} Therefore, the main objectives of treatment are cyst removal, splenic function preservation, and if possible, prevention of recurrences using minimally invasive approaches. However, when the spleen is mostly occupied by a cyst, as in our case, total splenectomy is required for cyst eradication.⁹ Furthermore, the recurrence of splenic cysts usually occurs in cases of true cyst that have not been completely removed.^{10–12} Thus, laparoscopic splenectomy is still useful, depending on the type, shape and location.

CA19-9 is a well-known tumor marker for pancreatic adenocarcinoma; however, benign diseases often show elevated serum CA19-9 levels. Approximately 30 cases of CA19-9-positive splenic cysts, most of epithelial origin (true cyst), have been reported.³ In our opinion, there are potentially many more cases that have not been evaluated for serum CA19-9 levels or other tumor markers. In a preoperative status, it is preferable to perform a needle biopsy to determine the cyst type; however, some instances of



Fig. 3. In the immunostaining analyses, the epithelium of the cyst was positive for CA19-9 antibody.

rupturing have been reported.¹³ In the present case, because CT findings revealed a hematoma, we did not perform a biopsy for it. If a needle biopsy cannot be performed for some reason, diagnosing the type of splenic cyst becomes difficult. The patient's history, such as trauma, is not very reliable and ultrasound and CT are not useful to determine the cyst type. In such cases, serum CA19-9 levels may aid in diagnosing the type of splenic cyst.

4. Conclusion

If possible, a spleen-preserving approach is preferable for treating splenic cysts. However, these approaches in true cyst cases increase the risk of recurrence and further surgical interventions. The surgical approach should be determined by the type, shape, location, and, most importantly, CA19-9 levels in the splenic cyst.

Conflict of interest statement

None.

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Consent

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

Author contributions

All authors have contributed significantly, and that all authors are in agreement with the content of the manuscript.

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