



Laparoscopic partial splenectomy for a giant splenic pseudocyst with elevated CA19-9: a case report

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Introduction and importance: Currently, there is a lack of reliable evidence on the management of splenic cysts, which are rare. Exploring the efficacy of laparoscopic partial splenectomy can aid in the accumulation of treatment-related evidence.

Case presentation: Here, we report the case of a 31-year-old female who was diagnosed with a giant splenic cyst with elevated serum CA19-9 and subsequently underwent laparoscopic partial splenectomy.

Clinical discussion: The effects of most treatment options for splenic cysts, including percutaneous aspiration and drainage, fenestration, and partial splenectomy, have not been confirmed by high-level evidence. With the development of minimally invasive surgery, laparoscopic partial splenectomy has drawn increasing attention. Additionally, the relationships between tumor markers and splenic cysts need to be further elucidated.

Conclusions: Laparoscopic partial splenectomy might be recommended for patients with splenic cysts, especially when the cysts are not completely covered by the splenic parenchyma.

Keywords: carbohydrate antigen 19-9, case report, laparoscopy, splenectomy, splenic cysts

Background

Splenic cysts are rare, with an incidence of ~1%^[1,2]. These cysts can be classified into parasitic and nonparasitic cysts according to their etiology or true and pseudocysts according to the presence or absence of a cellular epithelial lining. Nonparasitic splenic cysts (NPSCs), the majority of splenic cysts, can be the result of trauma, neoplasm, infection, and other causes^[3]. Additionally, elevations in the levels of certain tumor markers (CA19-9, CA125, CEA) have been reported in NPSCs, which increases the difficulty of distinguishing them from splenic neoplasms^[4,5]. Thus far, few researchers have reported the presence of idiopathic nonparasitic cysts, and many relevant studies are case reports or reviews. Here, we report the case of a patient with an idiopathic pseudocyst and a serum CA19-9 level elevation for whom laparoscopic partial

HIGHLIGHTS

- The study shows laparoscopic splenectomy's effectiveness in splenic cyst treatment.
- It explores CA19-9's utility in differentiating cysts from tumors, stressing marker surveillance.
- Cyst location and mass nature guide treatment to mitigate recurrence risks.
- Research on advanced techniques for splenic cyst surgery is still necessary.

splenectomy was successfully performed. This case has been reported in line with the SCARE 2023 criteria^[6].

Case presentation

A 31-year-old female patient with no complaints was admitted to our hospital for a splenic mass. Three years prior to presentation, a small (maximal diameter: 1.1 cm) splenic mass was identified through ultrasound examination and was suspected to be a splenic cyst. However, the mass had increased in size (maximal diameter: 11.2 cm) over the year prior to presentation. During this period, this patient had not received any medical treatment. She had not been to any endemic areas, nor had she been through a major trauma. Additionally, the patient had no history of cancer or genetic or chronic diseases.

No obvious abnormalities were found in the patient's physical examination upon admission. Her initial laboratory test results were unremarkable except for an increased serum CA19-9 level of 52.54 U/ml (normal: <27 U/ml). In addition, Epstein-Barr virus and hydatid test results were negative. Abdominal ultrasound reexamination showed an anechoic dark area in the upper pole of the spleen, ~8×11 cm in size, with a regular shape and no obvious blood flow inside. Further abdominal enhanced computerized tomography revealed that the size of the splenic upper

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This study conformed with the ethical guidelines of the Declaration of Helsinki and was approved by the Institutional Review Board of Our Hospital.

Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

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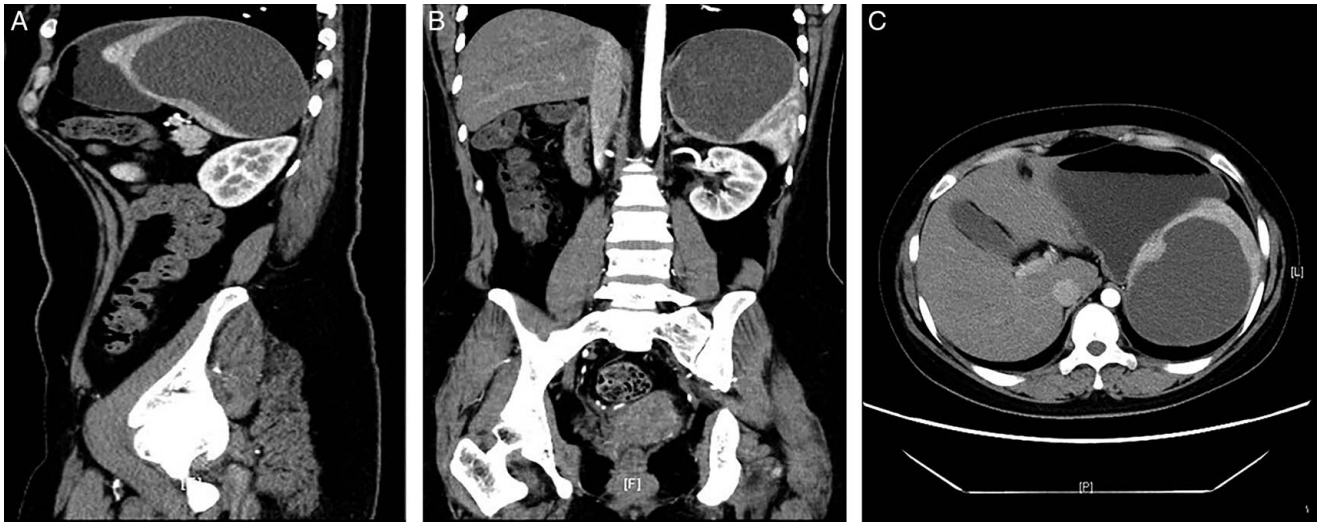


Figure 1. Enhanced abdominal computerized tomography showing a giant splenic cyst: (A) Sagittal view; (B) coronal view; (C) transverse view.

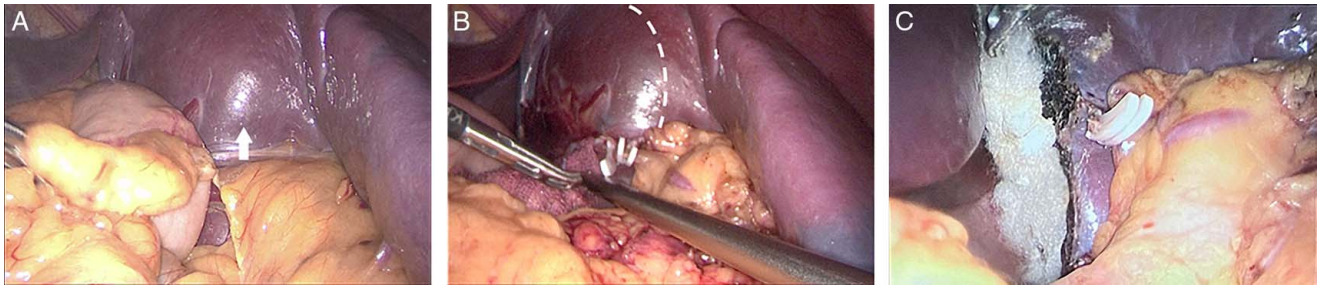


Figure 2. Intraoperative photos of laparoscopic partial splenectomy: (A) giant splenic cyst in the upper pole of the spleen (arrow); (B) necrotic boundary line after clamping the superior splenic blood vessel (dotted line); (C) hemostatic gauze coverage after cyst resection.

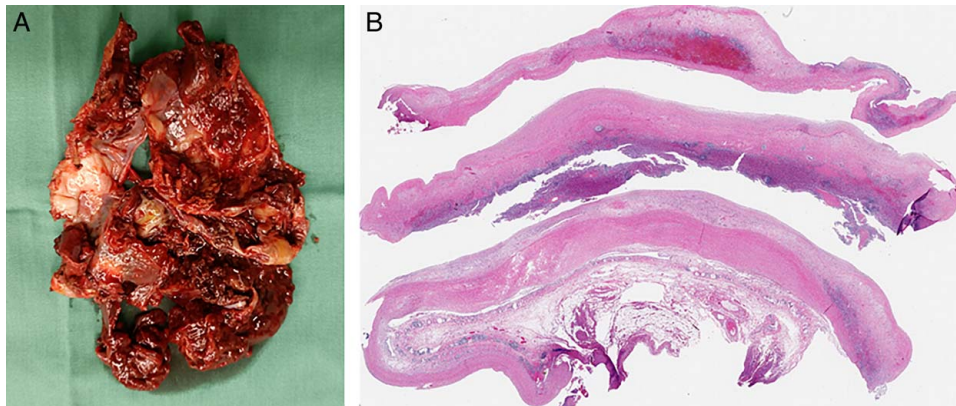


Figure 3. Operative specimen of splenic cyst. (A) The giant splenic cyst measured 10×8 cm with a wall thickness of about 0.2 cm; (B) Under the microscope, the structure of the cyst wall is mainly composed of fibrous connective tissue, abundant lymphocytes and histiocytes, dilated and bleeding small blood vessels, and calcified areas. No malignant tumor cells or epithelial cells were found.

pole mass was 8.4×11.2 cm, with irregular density and no significant enhancement after enhanced scanning (Fig. 1). Based on these results, we diagnosed this patient with an idiopathic non-parasitic splenic cyst.

To avoid complications such as cyst rupture or bleeding and to achieve maximal spleen preservation, we decided to perform laparoscopic partial splenectomy for this patient. Following the induction of general anesthesia, the patient was placed in a supine

Table 1
Characteristics of surgical methods for splenic cysts

Study/case reports ^a	Year	Cases	Procedure	Complications	Follow-up	Key findings
Akhan <i>et al.</i> ^[7]	1997–2015	24	Puncture-aspiration-injection-reaspiration/catheterization	7 recurrences	4–227 mo	Minimally invasive technique for preserving maximum tissue
Wang <i>et al.</i> ^[8]	2011–2013	6	Laparoscopic partial splenectomy	None reported	6 mo	
Lima <i>et al.</i> ^[9]	2010–2012	5	Laparoscopic partial splenectomy	None reported	10–18 mo	Innovative application of 3D virtual rendering
Uranues <i>et al.</i> ^[10]	1994–2005	16	Laparoscopic partial splenectomy	2 conversions and 2 pleural effusions	12 mo	
de la Villeon <i>et al.</i> ^[11]	2004–2012	6	Laparoscopic partial splenectomy	1 conversion	1 mo	
Cai <i>et al.</i> ^[12]	2010–2014	12	Laparoscopic partial splenectomy	None reported	5–60 mo	Laparoscopic approach should be attempted in nonparasitic cysts
Schier <i>et al.</i> ^[13]	1995–2005	14	Laparoscopic unroofing	9 recurrences	6–12 mo	Laparoscopic unroofing of true splenic cysts proved inadequate
Wu <i>et al.</i> ^[14]	1998–2004	6	Percutaneous drainage/laparoscopic fenestration	3 recurrences	2–5 mo	High rate of failure
Gianom <i>et al.</i> ^[15]	1996–2002	7	Open partial splenectomy/laparoscopic cyst wall unroofing	1 conversion and 1 recurrence	6–48 mo	Both tools are effective
Accinni <i>et al.</i> ^[16]	2008–2014	15	Ultrasound-guided percutaneous sclerosis	1 intraperitoneal fluid collection	3–79 mo	Valid and safe option for the pediatric population

^aStudies or case reports with a sample size of less than 5 or incomplete data were not included.

position. We initially introduced a 30° Storz laparoscope through a 10-mm port, situated 1 cm to the left of the umbilicus, to visualize the splenic cyst, which measured ~10 cm in its maximal diameter and was located at the upper pole. Subsequently, we inserted trocars 5, 12, and 5 mm in diameter at the following locations: 1 cm below the xiphoid process, 1 cm below the costal margin along the left midclavicular line, and 1 cm below the costal margin in the left anterior axillary line. The splenogastric ligaments and associated vasculature were meticulously dissected using an ultrasonic scalpel (Harmonic Scalpel, Ethicon Endo-Surgery), which facilitated the complete detachment of the splenic cyst from the surrounding adherent tissue. The superior arteriovenous pedicle of the spleen, which was situated on the surface of the cyst, was identified and ligated using Hemlock clamps. This maneuver resulted in the demarcation of the ischemic necrosis boundary around the cyst. Along this boundary, the cyst and a portion of the spleen were excised after complete aspiration of the sac fluid (Fig. 2). Hemostasis was achieved by employing an electrocoagulation rod, and hemostatic agents were applied to the surgical site. The drainage tube was then strategically positioned.

The operation time and blood loss were 120 min and 50 ml, respectively. This patient recovered well and could leave the bed one day after surgery. On the fifth day after surgery, the drainage tubes were removed. Reexamination blood tests indicated that the serum CA19-9 level had returned to within the normal range, but the platelet count had increased to 389×10^9 (normal: $125\text{--}350 \times 10^9$). Accordingly, oral aspirin (50 mg) was given every day. On postoperative day 7, this patient was discharged with no postoperative complications. Pathological examination confirmed that there were no epithelial cells on the cystic wall, indicating a spleen pseudocyst (Fig. 3). Two weeks postdischarge, the patient exhibited a normalization of the platelet count, prompting the cessation of oral medication. Subsequent ultrasonic or computed tomography imaging conducted at 3, 6, and 12 months postprocedure demonstrated no evidence of cyst recurrence. Furthermore, the patient reported no adverse symptoms or complaints during these follow-up intervals.

Discussion

With the development of surgical techniques, spleen-preserving approaches targeting splenic cysts are gradually gaining popularity. However, the effects of most treatment options, including percutaneous aspiration and drainage, fenestration, and partial splenectomy, have not been confirmed by high-level evidence (Table 1).

Percutaneous aspiration and drainage guided by imaging techniques are known to be minimally invasive and easy-to-perform procedures. However, the major concern for this technique may be its associated high recurrence rate, which is reported to be 29.1%, according to Akhan's report^[7]. In contrast, radical cyst resection can be achieved via splenectomy, and the assistance of laparoscopy helps reduce perioperative complications. Partial splenectomy is often preferred to total splenectomy because lesions can be radically resected and splenic function can be preserved. This advantage is the reason for the selection of this operation plan for our patient. However, surgeons should note that the cyst was mainly located in the upper pole of the spleen in this patient. If cysts are completely surrounded by splenic parenchyma, total splenectomy is recommended considering the risk of bleeding^[17]. Additionally, laparoscopic fenestration may be

applicable for most splenic cysts except splenic hydatid cysts, which require surgeons with high levels of experience and technical requirements^[18]. In recent decades, innovative methods such as 3D or robot-assisted surgery^[9,19], watchful waiting^[20], and indocyanine green dye^[11] have been proposed, but further validation of these methods in clinical practice is needed.

To differentiate splenic cysts from malignant tumors, we further tested the plasma levels of tumor markers and found only an increased CA19-9 level. Hoshino *et al.*^[21] and Madia *et al.*^[22] reported similar changes in serum tumor marker levels. However, abnormally high serum CA19-9 and CEA levels have been found in some other patients, especially those with ruptured cysts^[23]. Such changes have not been validated. One possible explanation may be that epithelial cells produce and release tumor markers into the cyst and blood. CA19-9 levels might be indicative of treatment effects on splenic epidermoid cysts since an obvious decrease in CA19-9 levels has been observed after treatment^[23]. Oddly, the splenic lesion of our patient was confirmed to be a pseudocyst without epithelial cells, but similar CA19-9 changes were still found. The significance of changes in tumor marker levels remains unknown, and these changes need to be validated. However, we believe that the use of tumor markers to differentiate tumors should always be the first priority.

Conclusion

Laparoscopic partial splenectomy is worthy of consideration when splenic cysts are not completely covered by the splenic parenchyma. However, the significance of changes in tumor marker (CA19-9, CEA, etc.) levels is still unclear, and these changes need to be validated. Large-scale and multicenter studies are urgently needed to solve the current dilemma of low-quality evidence regarding splenic cysts.

Ethical approval

The Ethics Committee of the People's Hospital of Suzhou New District (protocol code: 2023-124; date of approval: 2023/09/06).

Consent

Written informed consent was obtained from the patient involved in this study.

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Conflicts of interest disclosure

All the authors have no conflicts of interest related to the manuscript.

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Not applicable.

Guarantor

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Data availability statement

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Provenance and peer review

Not applicable.

References

- [1] Aggarwal RK, Mohanty BB, Prasad A. Laparoscopic splenic pseudocyst management using indocyanine green dye: an adjunct tool for better surgical outcome. *J Minim Access Surg* 2019;15:154–7.
- [2] Zvizdic Z, Karavdic K. Spleen-preserving surgery in treatment of large mesothelial splenic cyst in children—a case report and review of the literature. *Bosn J Basic Med Sci* 2013;13:126–8.
- [3] Jiang RD, Zhang ZL, Li T. Calcified cyst in the spleen. *JAMA Surg* 2016; 151:675–6.
- [4] Ballestri S, Lonardo A, Romagnoli D, *et al.* Primary lymphoma of the spleen mimicking simple benign cysts: contrast-enhanced ultrasonography and other imaging findings. *J Med Ultrason* (2001) 2015;42: 251–5.
- [5] Walz MK, Metz KA, Sastry M, *et al.* Benign mesothelial splenic cyst may cause high serum concentration of CA 19-9. *Eur J Surg* 1994;160: 389–91.
- [6] Sohrabi C, Mathew G, Maria N, *et al.* The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. *Int J Surg* 2023;109:1136–40.
- [7] Akhan O, Dagoglu-Kartal MG, Ciftci T, *et al.* Percutaneous treatment of non-parasitic splenic cysts: long-term results for single- versus multiple-session treatment. *Cardiovasc Intervent Radiol* 2017;40:1421–30.
- [8] Wang X, Wang M, Zhang H, *et al.* Laparoscopic partial splenectomy is safe and effective in patients with focal benign splenic lesion. *Surg Endosc* 2014;28:3273–8.
- [9] Lima M, Reinberg O, Ruggeri G, *et al.* 3D virtual rendering before laparoscopic partial splenectomy in children. *J Pediatr Surg* 2013;48: 1784–8.
- [10] Uranues S, Grossman D, Ludwig L, *et al.* Laparoscopic partial splenectomy. *Surg Endosc* 2007;21:57–60.
- [11] de la Villeon B, Zarzavadjian Le Bian A, Vuarnesson H, *et al.* Laparoscopic partial splenectomy: a technical tip. *Surg Endosc* 2015;29: 94–9.
- [12] Cai H, An Y, Wu D, *et al.* Laparoscopic partial splenectomy: a preferred method for select patients. *J Laparoendosc Adv Surg Tech A* 2016;26: 1010–4.
- [13] Schier F, Waag KL, Ure B. Laparoscopic unroofing of splenic cysts results in a high rate of recurrences. *J Pediatr Surg* 2007;42:1860–3.

- [14] Wu HM, Kortbeek JB. Management of splenic pseudocysts following trauma: a retrospective case series. *Am J Surg* 2006;191:631–4.
- [15] Gianom D, Wildisen A, Hotz T, *et al.* Open and laparoscopic treatment of nonparasitic splenic cysts. *Dig Surg* 2003;20:74–8.
- [16] Accinni A, Bertocchini A, Madafferi S, *et al.* Ultrasound-guided percutaneous sclerosis of congenital splenic cysts using ethyl alcohol 96% and minocycline hydrochloride 10%: a pediatric series. *J Pediatr Surg* 2016;51:1480–4.
- [17] Hansen MB, Moller AC. Splenic cysts. *Surg Laparosc Endosc Percutan Tech* 2004;14:316–22.
- [18] Vasilescu AM, Tarcoveanu E, Ciuntu B, *et al.* Laparoscopic approach for nonparasitic splenic cysts and splenic abscesses. *Ann Ital Chir* 2022;93:671–9.
- [19] Giulianotti PC, Daskalaki D, Gonzalez-Ciccarelli LF, *et al.* Robotic splenectomy with ex vivo bench surgery and hemi-spleen autotransplant: the first report. *J Robot Surg* 2017;11:243–6.
- [20] Di Lena E, Safa N, Rahman S, *et al.* Watchful waiting for large primary nonparasitic splenic cysts. *Can J Surg* 2023;66:E390–5.
- [21] Hoshino A, Nakamura Y, Suzuki H, *et al.* Giant epidermoid cyst of the spleen with elevated CA 19-9 production managed laparoscopically: report of a case. *J Nippon Med Sch* 2013;80:470–4.
- [22] Madia C, Lumachi F, Veroux M, *et al.* Giant splenic epithelial cyst with elevated serum markers CEA and CA 19-9 levels: an incidental association? *Anticancer Res* 2003;23(1B):773–6.
- [23] Imoto Y, Einama T, Fukumura M, *et al.* Laparoscopic fenestration for a large ruptured splenic cyst combined with an elevated serum carbohydrate antigen 19-9 level: a case report. *BMC Surg* 2019;19:58.