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CASE REPORT

A case of ruptured splenic cyst with elevated serum levels of CEA treated by laparoscopic unroofing

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Abstract
Splenic cysts are rare and tend to have elevated tumor markers, of which carbohydrate antigen (CA) 19-9 is the most frequently elevated. Therefore, splenic cysts with elevated serum carcinoembryonic antigen (CEA) levels and without CA19-9 elevation are extremely rare. A 26-year-old woman presented with sudden upper abdominal pain while sleeping. Contrast-enhanced computed tomography (CT) showed an 85-mm simple splenic cyst in the upper pole and a moderate amount of ascites around the spleen. The serum levels of CEA, but not CA19-9, were elevated. Spontaneous rupture of a splenic cyst was diagnosed. We performed elective laparoscopic unroofing. The histological findings revealed a stratified squamous epithelium on the inner surface of the cystic wall. On immunohistochemical examination, the squamous epithelium was found to be positive for cytokeratin (CK)7, negative for calretinin, and positive for CEA. The histological diagnosis was an epidermoid cyst. Three months after the operation, the elevated serum tumor marker levels of CEA were normalized. Splenic cysts with high levels of CEA and low levels of CA19-9 are extremely rare. Laparoscopic unroofing is a useful operative procedure for ruptured splenic cysts with elevated levels of serum tumor markers.

Keywords Epidermoid cyst · Carcinoembryonic antigen · Carbohydrate antigen 19-9 · Tumor markers · Laparoscopy

Introduction

Splenic cysts are still a rare entity, although clinical cases have been increasingly reported with recent advances in various diagnostic modalities. Epidermoid cysts of the spleen are categorized as a true splenic cyst lined with squamous epithelium and tend to have elevated tumor markers, such as carbohydrate antigen (CA) 19-9, carcinoembryonic antigen (CEA) and CA125. Among these markers, elevated serum levels of CA19-9 have been most frequently reported in several cases of epidermoid cysts [1–7]. However, it is extremely rare for an epidermoid cyst to cause elevated serum levels of CEA but low levels of CA19-9.

Most splenic cysts do not present with typical clinical symptoms and are usually found incidentally. Ruptured splenic cysts are rare [8–12], and sometimes, an emergency splenectomy is performed [10, 11]. Because the spleen has a variety of tasks in the defense system and is responsible for removing blood cells, splenectomy should be avoided when possible. In addition, advances in laparoscopy have enabled minimally invasive surgical treatment of splenic diseases. Although laparoscopic total splenectomy is an established surgical procedure, there are few reports on spleen-preserving surgery of ruptured splenic cysts [12]. We report a case of a spontaneously ruptured splenic cyst with elevated serum levels of CEA and CA125 and without CA19-9 elevation that was successfully treated with laparoscopic unroofing.

Case report

A 26-year-old Japanese woman presented with sudden upper abdominal pain while sleeping. She had never experienced this symptom, and there was no significant past medical and surgical history or history of trauma. On examination, the patient's pulse rate was 90 beats/min, her blood pressure was 107/70 mmHg, and her temperature was 36.5°. Her abdomen was distended, and she had diffuse abdominal tenderness and

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rebound tenderness in the upper abdomen. Laboratory tests revealed an elevated white blood cell count (10,200 cells/μl), but a normal C-reactive protein level (0.02 mg/dl) without anemia (Hb; 12.1 g/dl) (Table 1). Contrast-enhanced computed tomography (CT) showed an 85 × 75 mm simple splenic cyst in the upper pole without any extravasation and a moderate amount of ascites around the spleen, the liver surface and the Douglas pouch (Fig. 1a). The CT scan also revealed some small calcifications in a portion of the cystic wall (Fig. 1b). Spontaneous rupture of a splenic cyst was diagnosed. Because the pain was almost alleviated by using a nonsteroidal anti-inflammatory drug and after that, she had almost no symptoms, we decided to perform an elective surgery rather than an emergency surgery, and a further examination was performed. The serum levels of tumor markers CEA and CA125 were elevated (40.4 ng/ml and 887.7 U/ml), while that of CA19-9 was not (< 0.2 U/ml) (Table 1). Contrast-enhanced magnetic resonance imaging (MRI) showed that the cyst did not contain solid components or septal walls (Fig. 2). Abdominal ultrasonogram showed the splenic cyst contained homogeneously hypoechoic fluid with a few highly echogenic spots (Fig. 3). F-18 fluorodeoxyglucose positron emission tomography (FDG-PET)/CT showed no abnormal uptake in the splenic cyst or other sites (Fig. 4). Gynecological examination showed ascites in the Douglas pouch and no gynecological disease. Puncture of the Douglas pouch was performed, and the ascitic fluid was brown and turbid. The ascites fluid levels of CEA and CA 125 were as high as 1137.7 ng/ml and 12,690.9 U/ml, but that of CA19-9 was less than 2.0 U/ml. There was no cytological evidence of malignancy, and the bacterial culture result was negative. Because the possibility of malignancy was extremely rare according to these results, we decided not to perform a splenectomy but to administer spleen-preserving surgery by laparoscopy.

The patient was temporarily discharged from the hospital on day 4 after conservative treatment, and laparoscopic unroofing was performed 16 days after the onset. The patient was placed in a 45-degree right lateral decubitus position, and when necessary, the table was rolled to the left to place the patient in a nearly supine position. One transumbilical

Table 1 Laboratory data

Blood count	
White blood cell count	10,200 cells/μl
Hemoglobin	12.1 g/dl
Platelet count	28.3×10^4/μl
Serum chemistry	
Aspartate aminotransferase	14 U/l
Alanine aminotransferase	10 U/l
γ-Glutamyl transferase	8 U/l
Alkaline phosphatase	101 U/l
Total bilirubin	0.6 mg/dl
Lactate dehydrogenase	161 U/l
Amylase	82 U/l
Blood urea nitrogen	12 mg/dl
Creatinine	0.63 mg/dl
C-reactive protein	0.02 mg/dl
Tumor markers	
Carcinoembryonic antigen	40.4 ng/ml
Carbohydrate antigen 19-9	< 2.0 U/ml
Carbohydrate antigen 125	887.7 U/ml

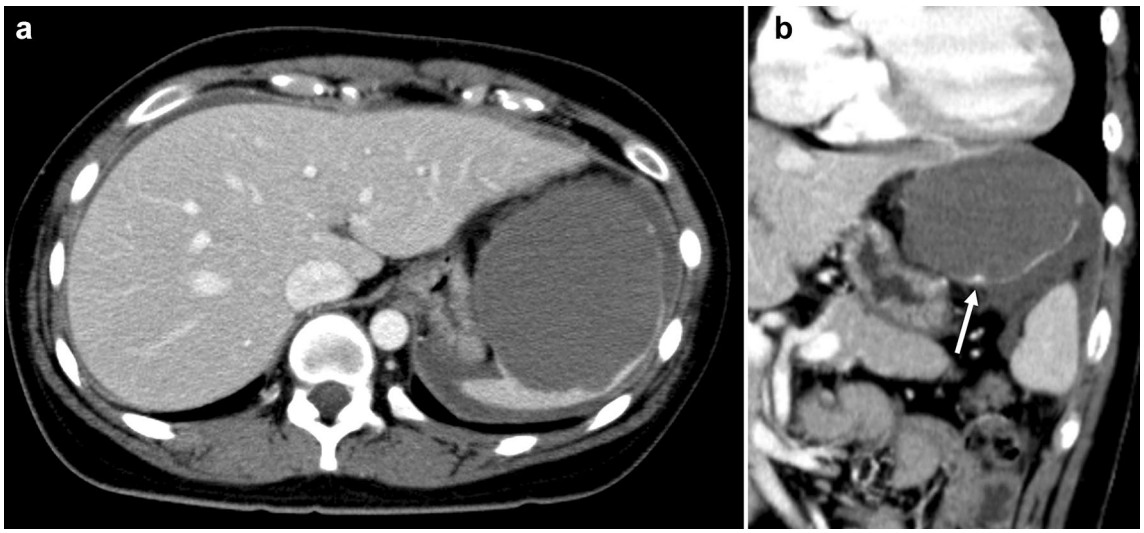

Fig. 1 Contrast-enhanced CT: **a** the axial plane shows an 85 × 75 mm simple splenic cyst in the upper pole and a moderate amount of ascites around the spleen and the liver surface. **b** The coronal plane shows the presence of some small calcifications in a portion of the cystic wall (arrow)

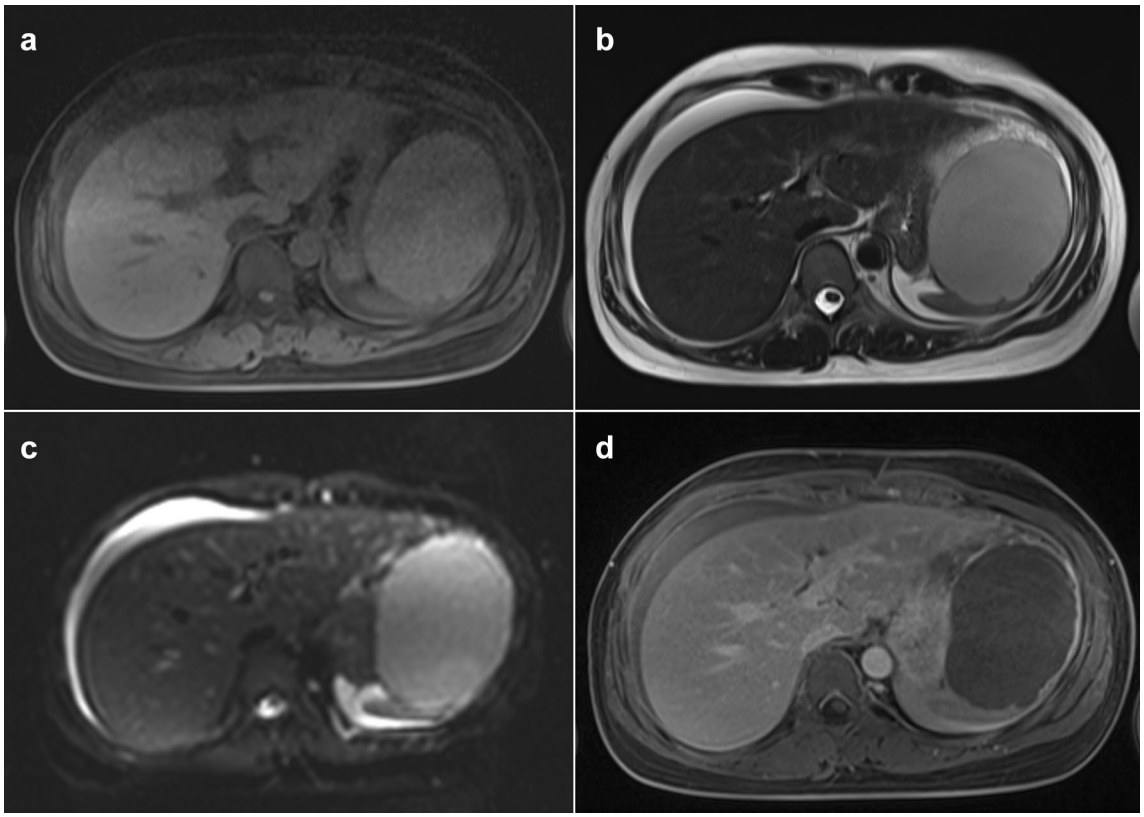

Fig. 2 Contrast-enhanced MRI: the cystic lesion is depicted as a slightly high-intensity lesion on the noncontrast T1-weighted image (**a**) and a slightly lower-intensity lesion than serous fluid on the T2-weighted image (**b**). There is no signal on the diffusion-weighted image (**c**) and no enhancement in the splenic cyst wall (**d** delayed phase)

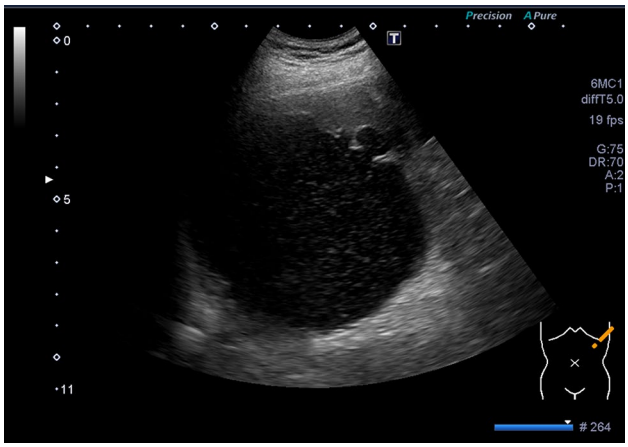

Fig. 3 Abdominal ultrasonogram showing the splenic cyst contained homogeneously hypoechoic fluid with a few highly echogenic spots

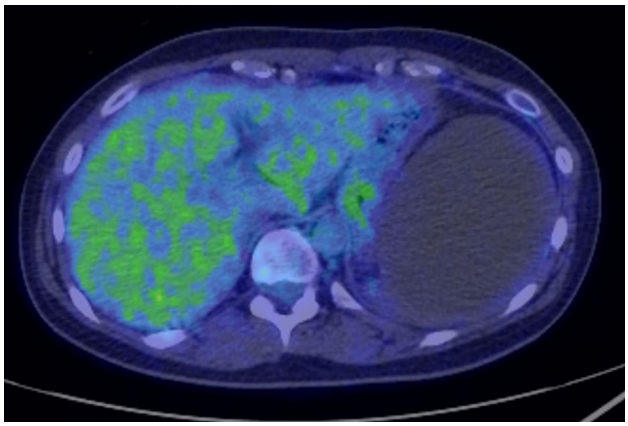

Fig. 4 F-18 fluorodeoxyglucose positron emission tomography (FDG-PET)/CT showing no abnormal uptake in the splenic cyst or other sites

access port for videolaparoscopy and three working trocars were inserted around the left hypochondrium. A Nathanson liver retractor was placed in the subxiphoid location, which retracted the stomach and left lateral segment of the liver to the right cranial portion. A yellowish-white cyst occupied the upper pole of the spleen, without ascites (Fig. 5). Intraoperative rapid cytological diagnosis of the fluid and pathological diagnosis of the cystic wall revealed no evidence of malignancy. The cyst wall was resected at the junction of the cyst and the spleen parenchyma using a LigaSure™ Vessel

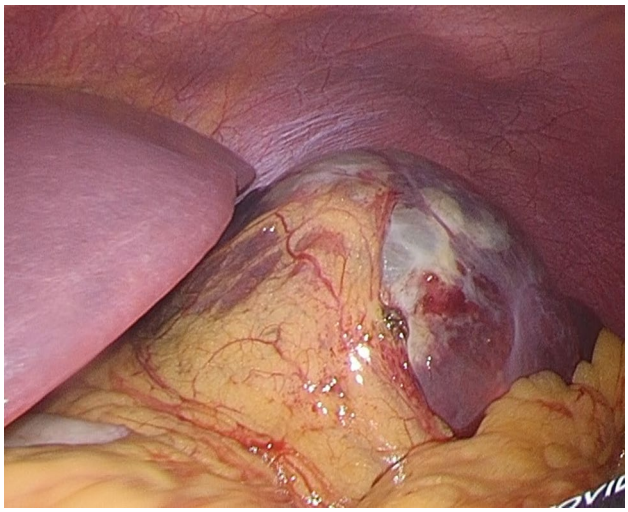

Fig. 5 Intraoperative photograph showing that the splenic cyst was yellowish-white and occupied the upper pole of the spleen, without ascites

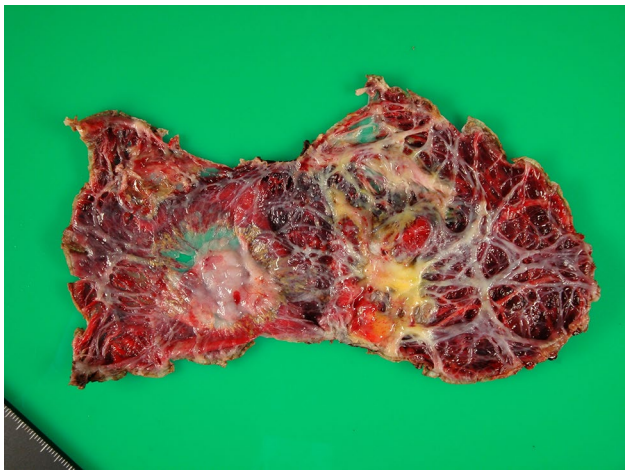

Fig. 7 Photograph of the surgical specimen showing that the interior of the cyst wall was white, glistening and partially thin and had prominent coarse fibrous trabeculations

Sealing System and was removed through the umbilical port site incision without difficulty. The residual cystic wall on the spleen was cauterized by a monopolar electrocautery with spray coagulation and a soft-coagulation system with a monopolar electrode (Fig. 6). The operative time was 2 h and 59 min, and the blood loss was 140 cc. The CEA and CA125 levels in the cystic fluid were high (4444.7 ng/ml and 63,026.3 U/ml, respectively), but the CA19-9 level was low (less than 2.0 U/ml).

Macroscopic findings revealed that the interior of the cyst wall was white, glistening and partially thin and had prominent coarse fibrous trabeculations (Fig. 7). The histological findings revealed stratified squamous epithelium on the inner surface of the cystic wall with a small amount of calcification. On immunohistochemical examination, the squamous epithelium was found to be positive for cytokeratin (CK)7, negative for calretinin, and positive for CEA (Fig. 8). The histological diagnosis was an epidermoid cyst.

The postoperative course was uneventful, and the patient was discharged on day 3 after surgery. Three months after the operation, the elevated serum tumor marker levels of CEA and CA125 were normalized (CEA, 1.5 ng/ml, and CA125, 21.3 U/ml) and were not elevated for up to 2 years after the operation. The patient remains healthy without recurrence, as confirmed by abdominal ultrasonography. Since the serum levels of CA19-9 were consistently less than measurable ($\leq$2.0 U/ml), we examined the Lewis blood group phenotypes, and the result was a Lewis-negative phenotype [Le (a− b−)].

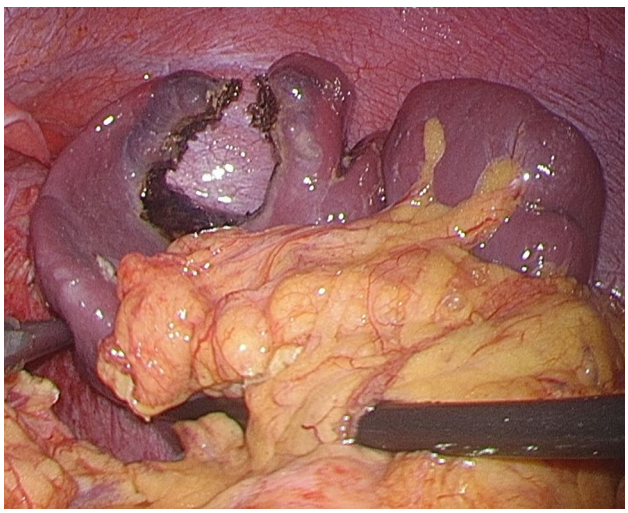

Fig. 6 Intraoperative photograph showing the final aspect of the residual cystic wall on the spleen, which was cauterized sufficiently

Discussion

Although splenic cysts are relatively rare, with the current use of abdominal imaging, the incidence of splenic cysts is thought to be increased [13]. The incidence of splenic cysts is higher in females than in males, and most cases of splenic cysts have occurred in young women or children [14]. Splenic cysts are classified as true cysts or pseudocysts. Primary true cysts of the spleen can be parasitic or nonparasitic, and epidermoid cysts account for 10% of all nonparasitic cysts [15], whereas most nonparasitic cysts are pseudocysts secondary to trauma. True cysts that are lined with squamous epithelium are classified as epidermoid cysts and thought to be congenital [16]. Epidermoid

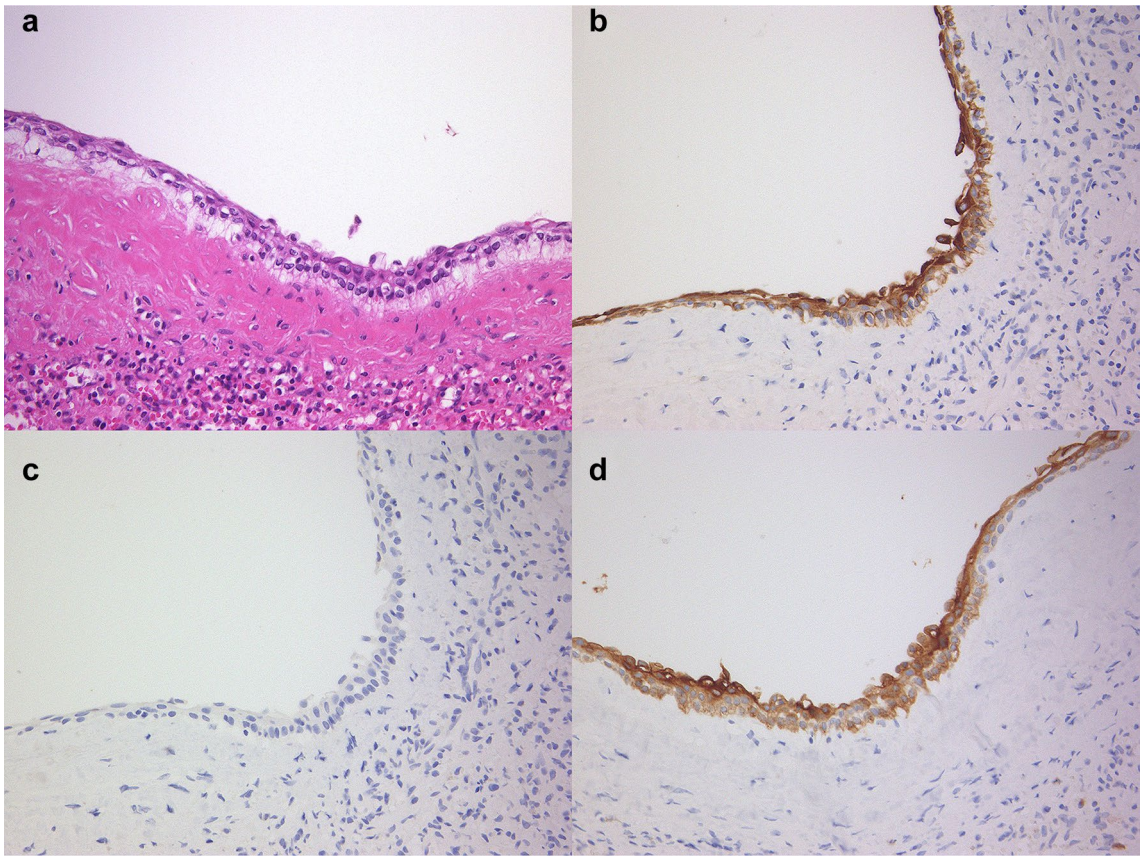

Fig. 8 Histopathological findings showing that the cyst wall was lined with stratified squamous epithelium (**a**). On immunohistochemical examination, the squamous epithelium was positive for CK7 (**b**), negative for calretinin (**c**), and positive for CEA (**d**)

cysts are positive for cytokeratin, but they are negative for calretinin, whereas mesothelial cysts are positive for both cytokeratin and calretinin [17]. It has been reported that patients with splenic epidermoid cysts have elevated serum levels of tumor markers, such as CA19-9, CEA and CA125 [1–5, 7, 10, 14, 18]. Most splenic cysts do not present with typical clinical symptoms and are usually found incidentally. Rarely, splenic cysts manifest with acute symptoms related to rupture, hemorrhage, or infection [8–12, 14]. Operative intervention is indicated for patients with symptomatic or large cysts.

As CA19-9, CEA and CA125 are well-known tumor markers for the diagnosis of digestive system and gynecologic cancers, there is some concern about splenic cysts with elevated tumor markers that might have malignant potential; in addition, ruptured splenic cysts cause severe peritonitis. Therefore, open splenectomy has been performed for patients with such lesions [3, 10]. The present case, however, showed the possibility that spleen-preserving surgery is indicated for ruptured splenic cysts with high serum levels of tumor markers.

Epidermoid cysts that have squamous epithelium on the inner surface of the cystic wall tend to have elevated tumor markers. There have been approximately 30 published cases of splenic cysts producing CA19-9 [6]; however, there is no case in the literature that had splenic cysts with elevated serum levels of CEA but low levels of CA19-9. The mechanism underlying a splenic cyst with both high serum levels of CEA and low levels of CA19-9 has not yet been elucidated. However, we hypothesized and verified that such patients have a Lewis blood group-negative phenotype [Le (a− b−)] because the serum levels of CA19-9 are consistently less than measurable ($\leq$2.0 U/ml). The frequency of this phenotype is approximately 5–10% among Japanese people [19], and there are some reports indicating that 94.7–100% of patients with Lewis-negative phenotypes have undetectably low CA 19-9 values because these individuals cannot synthesize CA19-9 [19–21]. Therefore, the two patients have similar histologic features of stratified squamous epithelium, which may be the same disease as CA19-9-producing splenic cysts.

Some patients with CA19-9-producing splenic cysts also have a high concentration of CEA in the cystic fluid, while its serum levels are normal [7, 14, 22]. Tumor markers are hypothesized to be secreted into the intracystic fluid by inner epithelial cells originating from the cyst

[10]. Positivity for anti-CEA and CA19-9 antibodies in the inner epithelium supports CEA and CA19-9 production. In patients with ruptured splenic cysts, blood analyses as well as cystic fluid analyses show elevated levels of CEA [10, 18]. In cases with no rupture, only CA19-9 enters the blood through the surrounding splenic tissues and CEA does not, and therefore, only serum CA 19-9 is elevated, and CEA does not. On the other hand, in ruptured cases, it would appear that CEA contained in the cystic fluid is also absorbed into the blood via the peritoneum together with CA19-9; consequently, the serum CEA is also elevated [10]. In the present case, it might be that the ruptured splenic cyst caused high serum levels of CEA and the Lewis blood group-negative phenotype that did not express CA19-9 caused low levels of CA19-9.

Traditionally, splenectomy is considered the treatment of choice for splenic cysts with elevated tumor markers [3, 7, 10, 14, 18]. This might be because splenic cysts with elevated tumor markers are thought to have the possibility of malignancy and should be resected completely, avoiding intraoperative leakage of cystic fluid. On the other hand, it is possible that serious early and late complications, such as overwhelming postsplenectomy infection (OPSI), reactive thrombocytosis and portal or splenic vein thrombosis, are occasionally caused by splenectomy [23]. OPSI is the most common fatal late complication of splenectomy [24]. Infection may occur at any time after splenectomy [3]. Although the incidence of OPSI in adults is low, mortality rates from sepsis that are as high as 50% have been reported [23]. Based on these observations, efforts should be made to preserve as much splenic parenchyma as possible to prevent such fatal complications, especially in younger patients who have a high proportion of splenic cysts. In addition, few cases of malignant splenic cysts have been reported worldwide [25, 26], and findings from these images might reveal not classic but rather unique characteristics. Even if the tumor marker levels are high, such cysts are less likely to be malignant. Therefore, it is prudent to perform spleen-preserving surgery as much as possible, based on excluding the possibility of malignancy by various diagnostic modalities.

Reported cases of ruptured epidermoid cysts of the spleen with elevated tumor markers are extremely rare, and only 4 cases have been reported, including the present case (Table 2) [3, 10, 18]. Two patients who had tests of ascites fluid showed elevated ascites levels of CEA, and another patient exhibited elevated levels of CEA in the cystic fluid. Despite elevated levels of tumor markers, no malignancy was detected in these patients, and the serum levels of the tumor markers were reduced after resection. There are no reports of local recurrence, dissemination or persistently elevated levels of tumor markers after resection.

Advances in laparoscopy have enabled the minimally invasive surgical treatment of splenic diseases. Laparoscopic splenectomy can be performed easily and safely and is the standard surgical procedure performed in most centers worldwide. Open splenectomy is still indicated in abdominal trauma, and in cases where the spleen size does not permit safe laparoscopic resection. There are some reported cases of splenic cysts resected by laparoscopic partial resection [12, 27–29]. To prevent recurrence after the treatment of primary splenic cysts, a partial splenic resection seems to be highly efficacious. In partial splenic resection, the splenic cyst wall is completely resected together with the part of the spleen where the lesion was attached. Resection of splenic tissue with electrocautery and ultrasonic scissors is, however, generally more technically demanding and time consuming because cutting the spleen causes considerable bleeding that increases the risk of conversion to open surgery and complications, such as excessive intraoperative and postoperative hemorrhage requiring transfusion. Therefore, it would be difficult for anyone to perform this procedure easily and safely. The technique of laparoscopic unroofing is safe and easy for laparoscopic surgeons and can be advantageous in terms of tissue sparing. The first laparoscopic procedure of unroofing a splenic cyst was reported by Salky et al. [30]. Laparoscopic unroofing is a spleen-preserving procedure that creates a splenic cyst-peritoneal window for drainage. At the present time, names such as “deroofing”, “partial resection”, “fenestration” or “decapsulation” are

Table 2 Reported cases of ruptured epidermoid cysts of the spleen with elevated tumor markers

No.	Author	Year	Age (years)	Sex	Operation	Serum			Cystic fluid			Ascites		
						CA19-9	CEA	CA125	CA19-9	CEA	CA125	CA19-9	CEA	CA125
1	Matsubayashi [18]	2001	36	F	LSP	**> 24,000**	**8.3**	ND	**> 24,000**	**3417**	ND	ND	ND	ND
2	Kubo [3]	2005	15	M	OSP	**214**	ND	ND	**> 5000**	**383**	**3041.1**	ND	ND	ND
3	Inokuma [10]	2010	20	F	OSP	**43,000**	**135**	**1470**	ND	ND	ND	ND	**7700**	ND
4	Our case	2018	26	F	LUR	< 0.2	**40.4**	**887**	< 2.0	**4444**	**63,026**	< 2.0	**1137**	**12,690**

Abnormal values are in bold

CA19-9 carbohydrate antigen 19-9, *CEA* carcinoembryonic antigen, *CA125* carbohydrate antigen 125, *M* male, *F* female, *LSP* laparoscopic splenectomy, *OSP* open splenectomy, *LUR* laparoscopic unroofing, *ND* not described

used synonymously to describe the operation. However, "decapsulation" is a misnomer because the spleen is not decapsulated, leaving the attachment part of the cyst wall on the spleen [27]. Recurrence is a problem associated with this procedure [17, 31, 32]. In the case of true cysts, cystic fluid is produced from the epithelial cells of the cystic wall, which might be a cause of recurrence and the production of tumor markers. Therefore, it is important to divide the cyst wall at the junction of the cyst and the spleen parenchyma to resect the cyst wall as much as possible and cauterize the residual cystic wall lumen on the spleen sufficiently [32, 33]. In consideration of this fact, we cautiously cauterized the residual cyst wall of the spleen in this case, and there has been no sign of recurrence, but the long-term follow-up evaluation after this procedure is unclear.

Since peritonitis from ruptured epidermoid cysts is not fatal, an elective laparoscopic operation is feasible for such patients. Although care should be taken to avoid intraoperative dissemination of the cystic contents, it was generally acceptable in this case because the cyst already ruptured at the outset. In addition, infectious disease and malignancy were found to be negative by the preoperative test results, such as FDG-PET/CT, bacterial culture analysis and cytological diagnosis of the ascites. Laparoscopic unroofing, which can preserve splenic function and offers cosmetic benefits, is a good indication and considered to be a useful operative procedure for splenic cysts with elevated levels of tumor markers, even if these cysts are ruptured.

Conclusions

Splenic cysts with high levels of CEA and low levels of CA19-9 are extremely rare. They may, however, represent the same disease as CA19-9-producing splenic cysts. Laparoscopic unroofing is a useful operative procedure for splenic cysts with elevated levels of serum tumor markers.

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Compliance with ethical standards

Conflict of interest Masataka Okuno, Norihiro Yuasa, Eiji Takeuchi, Yasutomo Goto, Hideo Miyake, Hidemasa Nagai, Yuichiro Yoshioka and Kanji Miyata declare that they have no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from the patient for the publication of this report.

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