Case report

Spontaneous rupture of a non-parasitic splenic cyst

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SUMMARY

Rupture of a non-parasitic splenic cyst is a rare but possibly dangerous complication with 21 cases described so far. We present a 46-year-old woman who presented with acute abdominal pain and was diagnosed with a spontaneous ruptured splenic cyst that was successfully treated by laparoscopic splenectomy. Histological examination showed characteristics corresponding with a non-parasitic congenital cyst that had lost its epithelial lining. Several treatment options can be considered for splenic cysts, depending on size and location. In case of rupture, the clinical condition of the patient should be taken into account.

BACKGROUND

Although the non-parasitic splenic cyst is an uncommon disorder, over 1000 cases have been reported throughout the years.¹ As patients may remain asymptomatic, splenic cysts can be incidentally diagnosed. More often, splenic cysts are diagnosed when they become symptomatic and patients present themselves with a feeling of discomfort in the left upper quadrant of the abdomen.^{1 2} The classification of non-parasitic splenic cysts has been discussed often. The most recently suggested division in primary and secondary seems to be most justified: primary cysts include congenital and neoplastic cysts, and secondary cysts include traumatic and necrotic cysts (table 1).³ Based on the cystic lining, congenital cysts can be further divided into mesothelial and metaplastic mesothelial cysts, the latter including epidermoid cysts. However, there are indications that these congenital cysts can lose their epithelial lining.⁴ This has resulted that congenital splenic cysts were incorrectly diagnosed as post-traumatic cysts in the past. Neoplastic cysts are subdivided in angiomata and dermoid cysts. Although infrequently encountered, the potential occurrence of complications should be taken into account.^{1 5 6} Rupture of a non-parasitic splenic cyst is a rare but possibly dangerous complication, with risk for developing infection (eg, peritonitis, abscess, empyema) or bleeding (haemoperitoneum).⁷ So far, 21 cases have been described who have undergone different surgical treatments suggesting there is no consensus yet. We present a female adult with a ruptured, presumably congenital, splenic cyst who was successfully treated with laparoscopic splenectomy.

CASE PRESENTATION

A 46-year-old woman presented at our emergency department with acute pain in the left abdominal upper quadrant, the lower left chest and the left shoulder without recollection of a preceding trauma. Other symptoms she had were nausea, pale-ness and perspiration. The patient had a medical history of an asymptomatic splenic cyst of 11 cm in diameter, which was an incidental finding during a familial screening for colorectal cancer elsewhere in 2013. There were no other family members with splenic cysts and she did not use any medication. Physical examination revealed a painful left abdominal upper quadrant and a palpable mass beneath the costal bones on the left side. There were no other remarkable symptoms and vital signs were within normal limits. The pain had decreased since it started (Visual Analogue Score (VAS) 8) and was still decreasing at the moment of arrival in the hospital (VAS 4).

INVESTIGATIONS

Blood tests did not show any significant increased infection parameters (leucocytes $10.8 \times 10^9/L$ and C-reactive protein (CRP) $<1 \text{ mg/L}$). Tumour markers were not determined as these have no clinical significance for the treatment of splenic cysts.⁸ Ultrasound (US) of the abdomen showed a splenic cyst sized $10.0 \times 8.8 \times 10.5 \text{ cm}$ surrounded by free fluid, from which 12 cc was punctured. The aspirate was turbid and yellow, and consisted of benign cells and inflammatory debris. CT scan of the abdomen confirmed the presence of a splenic cyst, located in the hilum, and it revealed a rupture of the lining of the cyst on the dorsal inferior border (figure 1). The somewhat decreased round shape of the cyst suggested that the pressure on the cystic wall had declined, meaning that fluid was or had been slowly oozing out of the cyst. The CT did not show any other abnormalities. Because of the clinical condition (normal vital signs and decreasing pain), the mild blood test results, the lack of blood in the fluid, and the CT findings suggesting a small rupture that would probably close again, conservative treatment was chosen in agreement with the patient, also considering her preferences. She was still admitted for observation and blood tests were performed the following day, which showed slightly increased infection parameters (leucocytes $12.5 \times 10^9/L$ and CRP 24 mg/L). However, the pain had decreased further and she was discharged in good clinical condition. Blood tests 2 days after discharge showed decreased infection parameters (leucocytes $8.1 \times 10^9/L$ and CRP 11 mg/L). One and half month later, the pain was slowly coming up

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Table 1 Classification of non-parasitic cysts³

Primary	Secondary
Congenital	Traumatic
Mesothelial	Necrotic
Metaplastic	
Transitional	
Stratified squamous	
Neoplastic	
Angioma	
Haemangioma	
Lymphangioma	
Dermoid	

again and a CT was performed again showing that the cyst had slightly increased in size. There was no rupture anymore and no free fluid in the abdomen. Consequently, she was planned for surgical treatment. The aim was to perform laparoscopic marsupialisation, but other scenarios (partial and total splenectomy) were also discussed with the patient in case marsupialisation would fail.

TREATMENT

Three months after initial presentation, laparoscopic splenectomy was performed under general anaesthesia and after antimicrobial prophylaxis by cefazolin and metronidazole. Due to the intra-abdominal view in combination with the location

Figure 1 Sagittal view of CT-scan with rupture indicated. **Figure 2** Upper: wall of the cyst (H&E, 20x); lower: lymphocytes, histiocytes, plasma cells and eosinophils. No epithelial lining is present (H&E, 400x).

and size of the cyst, marsupialisation and partial splenectomy were not considered as appropriate options anymore. The large spleen with splenic cyst was mobilised with a harmonic scalpel (Ultracision, Ethicon Endo-Surgery, Cincinnati, Ohio, USA), during which the gastrosplenic ligament and the vasa brevia were ligated. The splenic hilum was revealed and inferiorly, the splenocolic ligament was cut. One by one, the splenic artery and vein were exposed and cut after applying Hem-o-lok clips (Teleflex, Morrisville, North Carolina, USA). The spleen together with the intact cyst was further mobilised, placed in an Endo Bag (Medtronic, Minneapolis, Minnesota, USA) and retrieved through the extended left inferior incision hole after it was manually fragmented in the abdomen. Finally, the cavity was flushed with NaCl 0.9%, haemostasis was achieved and the incision wounds were closed. The laparoscopic splenectomy was completed without any perioperative complications. There was a blood loss of 400 mL.

OUTCOME AND FOLLOW-UP

The resected formalin-fixed fragments of the cyst and parenchyma had a volume of 136 mL and one fragment showed a diameter of 7 cm. The thickness of the wall of the cyst varied from 2 to 4 cm and there were no papillary structures on the inside of the wall, although it did show connective tissue. Although the microscopic findings were somewhat atypical (figure 2), the absence of an epithelial lining seemed to be matching the most with a congenital cyst that had lost its epithelial lining, or with

a benign secondary cyst, presumably traumatic or secondary to an infection of the spleen. As a subclinical course of an infection of the spleen is unlikely, it was probably not a cyst secondary to infection. The patient did not recall any trauma in the past. This does not necessarily exclude the possibility of a traumatic cyst, but it makes a congenital cyst more likely.

One day postoperatively, the patient was discharged with normal vital signs and some slight pain (VAS 3 out of 10). She was scheduled for administration of pneumococcal conjugate vaccine (PCV13), pneumococcal polysaccharide vaccine (PPSV23), haemophilus influenzae type B vaccine (Hib) and meningococcal C conjugate vaccine (Men-C). As the initial plan was to perform spleen preserving surgery, these vaccinations were unfortunately not administered preoperatively. Due to a penicillin allergy, clarithromycin therapy was started for 2 years. Six months after surgery, the patient was seen for follow-up and she presented with a painless swelling in the left upper quadrant at the location of the incision of the operation. Echo of the abdomen revealed a hernia cicatricialis of 2 cm with omental fat bulging through. To avoid that any complaints would arise, the hernia was corrected successfully by surgery. There was no specific explanation for this complication. The further postoperative course was uneventful and one and half a year after the initial operation she did not have any complaints.

DISCUSSION

Although non-parasitic splenic cysts are uncommon, more than 1000 case reports have been published until now. However, as ruptured cysts are even more uncommon, we concluded that 20 studies have been published addressing 21 cases with ruptured non-parasitic splenic cysts, the oldest study being published in 1960 (table 2).^{8–26} Age varies from 6 to 46 years old with our study presenting the oldest patient. All cases presented with abdominal pain which was sometimes accompanied by left shoulder pain, nausea or vomiting. Spontaneous as well as traumatic ruptures have been described. Sizes of the cysts varied from 4 to 14 cm and most were epidermoid cysts, which are the most common non-parasitic splenic cysts.²⁰ Accordingly, this type is also the most described ruptured cyst in literature. In 2007, Mirilas *et al* reassessed different classifications and suggested an adjusted classification based on pathogenesis and macroscopic and clinical characteristics (table 1).³ Holding on to this classification while keeping in mind that congenital cysts can lose their epithelial lining and that there was no trauma or infection in the past, we concluded that this most probably concerned a congenital cyst.⁴ Patients with a ruptured splenic cyst have risks of encountering dangerous consequences of this complication such as haemoperitoneum, peritonitis, abscess, anaphylactic shock or empyema.⁷ For our patient, because of the clinical and radiological findings together with the blood test results, we did not expect any of these dangers. We decided that waiting was allowed and admitted her for observation. It was until one and half month later that she became symptomatic again. As there were no signs of a rupture anymore, she was planned for surgery on the elective programme.

Various treatment options for splenic cysts have been described, including conservative treatment, percutaneous drainage, fenestration, marsupialisation partial splenectomy and total splenectomy.⁷

Several studies have suggested a conservative treatment strategy for non-parasitic splenic cysts with a diameter less than 5 cm although this statement is still under discussion as the evidence for this arbitrary cut-off point is rather poor.¹¹⁶ In fact,

a more recent study reviewed the literature and recommended that, regardless the size, asymptomatic splenic cysts with benign imaging characteristics should generally be managed conservatively.²⁷ This strategy is also used for hepatic cysts. As our case was asymptomatic until presentation, she was not offered surgical treatment earlier.

When a splenic cyst becomes symptomatic, surgical treatment is indicated. Percutaneous drainage (with or without sclerotherapy) is not recommended as it brings high recurrence rates, but it can still be used as a diagnostic tool by examining whether the symptoms resolve after aspiration.^{7,27} When surgery is performed, spleen preserving techniques are preferred as preservation of the spleen prevents the need for vaccinations and longtime use of antibiotics. However, it depends on the size and exact location of the cyst whether preservation is possible.

Superficially located cysts can be treated by fenestration or marsupialisation, having the advantage of leaving all the splenic parenchyma intact. Laparoscopic approach is preferred because of lower risks for complications, faster recovery, less pain and better cosmetic outcome, but an open approach can also be performed.⁷ By fenestration, a part of the wall of the cyst is removed, creating an opening to the peritoneum, after which a piece of omentum is attached to the surface of the spleen to minimise the risk for recurrence of the cyst. Studies have described that a sufficient part should be excised to prevent recurrence although there is no evidence for this.²⁸ While there is limited data of recurrence rates of splenic cysts, the recurrence rate for hepatic cysts is 9.6% after laparoscopic fenestration.²⁹ For marsupialisation, which involves removal of a part of the cyst after which the cut edges are sutured, there is no risk for recurrence and therefore it is the treatment of choice for superficial cysts.⁷ Both fenestration and marsupialisation are relatively simple procedures that have a short duration.

Splenic cysts located in the superior or inferior pole lend themselves for partial splenectomy. As estimated, 25% of the splenic parenchyma is at least needed to retain immunologic protection.⁷ Generally, blood supply of the spleen is provided by two or three branches of the splenic artery, supplying both poles and the intermediate part of the spleen. The arterial branches and corresponding veins leading to parenchyma where the cyst is situated should be ligated to avoid bleeding and to create demarcation of the ischaemic parenchyma that is to be resected. The spleen can be mobilised by ligating the vasa brevia (short gastric arteries), the gastrosplenic ligament and, if needed, the splenocolic ligament. Similar to fenestration and marsupialisation, partial splenectomy can be performed by open or laparoscopic approach.

For larger cysts affecting both poles, cysts located in the hilum or deeply in the parenchyma and polycystic spleens, spleen preservation is not achievable and total splenectomy is indicated, preferably by laparoscopy.²⁸ Also when other treatments fail or when uncontrollable bleeding occurs, total splenectomy should be performed.

As for the ruptured splenic cyst, the preferred treatment also depends on the clinical condition of the patient. In contrast to previous cases with a ruptured splenic cyst, we initially chose a conservative treatment strategy as the circumstances allowed this. In emergency setting with a clinically instable patient however, surgical treatment should be performed quickly. Spleen preserving surgery is theoretically possible depending on exact location and size of the cyst, but it will cost more time and it may be technically more demanding, which may be undesirable in acute setting. Depending on what is encountered, the operator

Table 2 Case reports of ruptured non-parasitic splenic cysts

First author	Year	Age	Gender	Presentation at hospital	Size (cm)	Treatment	Pathology	Complications	Free fluid
Coleman ⁹	1960	17	Female	Abdominal pain, vomiting	10	Total splenectomy, laparotomy	Epidermoid cyst	None	Large amount of blood
Browne ¹⁰	1963	9	Male	Left upper quadrant pain, nausea	12	Total splenectomy, laparotomy	Epidermoid cyst	Acute urine retention	None
Lippitt ¹¹	1967	10	Male	Abdominal pain, fever	7	Total splenectomy, laparotomy	Epidermoid cyst	Discharge with drain	NR
Watkins ¹²	1970	14	Female	Abdominal pain	6	Total splenectomy	Epidermoid cyst	None	Existence
Spence ¹³	1983	14	Male	Abdominal pain	11×7	Total splenectomy, laparotomy	Epithelial cyst	None	1.5 L of blood
Panossian ¹⁴	1990	20	Male	Abdominal pain	5.5×4.0	Total splenectomy	Epidermoid cyst	None	500 mL pus
Rathaus ¹⁵	1991	15	Female	Abdominal pain, vomiting	12	Total splenectomy, laparotomy	Epidermoid cyst	None	Large quantity of fluid
Musy ¹⁶	1992	15	Male	Abdominal pain, left shoulder pain	10	Marsupialisation	Post-traumatic cyst	None	1 L of blood
Musy ¹⁶	1992	13	Male	Upper quadrant irritation	8	Digitoclasy	Epidermoid cyst	None	500 mL blood
Spencer ¹⁷	1996	9	Male	Abdominal pain, vomiting	7	Fenestration, laparoscopic	Epidermoid cyst	None	None
Lam ¹⁸	1998	34	Male	Epigastric pain, left shoulder pain	5	Total splenectomy, laparotomy	Mesothelial cyst	None	600 mL blood
Matsubayashi ¹⁹	2001	36	Female	Left upper quadrant pain	14×9×7	Total splenectomy, laparotomy	Epidermoid cyst	None	Existence
Kiriakopoulos ²⁰	2005	32	Female	Left upper quadrant pain	9×6	Total splenectomy, laparoscopic	Epidermoid cyst	NR	Significant amount of fluid
Karasakalides ²¹	2006	35	Female	Upper abdominal pain	4	Total splenectomy, conversion	True splenic cyst	None	Small amount of blood
Kubo ²²	2006	15	Male	Abdominal pain	13.5×11.5	Total splenectomy	Epidermoid cyst	None	Existence, resolved
Inokuma ²³	2010	20	Female	Abdominal pain, left shoulder pain	11	Total splenectomy, laparotomy	Epidermoid cyst	None	Purulent fluid
Papadopoulos ²⁴	2010	21	Female	Tender left upper quadrant	12.2×11×10.2	Partial splenectomy, laparotomy	Epithelial cyst	None	NR
Tassopoulos ²⁵	2017	6	Female	Abdominal pain	7.1×6.2×6	Partial splenectomy, laparoscopic	Congenital cyst	None	Moderate to large amount of fluid
Okuno ⁸	2019	26	Female	Upper abdominal pain	8.5×7.5	Fenestration, laparoscopic	Epidermoid cyst	None	Turbid and brown fluid
Imoto ²⁶	2019	23	Female	Abdominal pain, vomiting	12×12×8	Fenestration, laparoscopic	Epidermoid cyst	None	Existence
Res (our case)	2019	46	Female	Left upper quadrant pain, nausea	10.0×8.8×10.5	Total splenectomy, laparoscopic	Congenital cyst	Hernia cicatricalis	Turbid an yellow fluid

NR, not reported.

has to weigh the benefits against the disadvantages to make the right decision.

Patient's perspective

Several years ago, I was diagnosed with a splenic cyst by coincidence. It never bothered me and the doctor from another hospital told me it was benign and ensured me it was harmless for now. Some years later, I suddenly became sick at work: I was sweating and pain was coming up in my left chest, left shoulder and abdomen. I visited the emergency department and after a scan it turned out that my splenic cyst was ruptured and that there was free fluid in my abdomen. For one night, I was admitted for observation and the day after I could go home. One and half a month later, I started to feel the pain again. The doctors decided to plan an operation and a total splenectomy was performed. After this operation, I recovered quickly. I received several vaccinations and the doctors told me to be aware of infections in the future. For 2 years, I had to take antibiotics. Except for that, I was not limited in my daily functioning. Six months passed when I noticed a swelling located at the scar of the incision of the operation. This appeared to be an incisional hernia and was corrected surgically. Nowadays, I do not have any complaints anymore.

Learning points

- Rupture of a non-parasitic splenic cyst is a rare complication and may be life-threatening.
- Symptoms can also be mild, in which case initial conservative treatment can sometimes be allowed.
- Spleen preserving surgery is an option depending on location and size of the splenic cyst.
- Our case was successfully treated by laparoscopic total splenectomy with full recovery.

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