

Epithelial splenic cysts and life-threatening splenic rupture

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Rezumat

Chisturile epiteliale splenice și ruptura splenică care pune viața în pericol

Premize/Scopul lucrării: Chisturile primitive splenice sunt relativ rare și sunt de cele mai multe ori clasificate drept chisturi epiteliale, acestea fiind mai frecvente la tineri. În majoritatea cazurilor sunt asimptomatice fiind descoperite incidental la examenele imagistice sau în timpul laparotomiei. Raportăm două cazuri rare de pacienți cu chisturi epiteliale splenice descoperite incidental în timpul laparotomiei de urgență pentru ruptură splenică.

Material și Metodă: Primul pacient este un bărbat de 70 ani care s-a prezentat cu simptome de abdomen acut după un acces de tuse în timpul unui episod de astm. Laparotomia de urgență a descoperit ruptură de splină și hemoragie intraperitoneală. S-a practicat splenectomie iar examenul histopatologic a evidențiat ruptură splenică cu un chist epitelial. Al doilea pacient este un bărbat de 19 ani care a fost transferat în Departamentul de Urgență în șoc hipovolemic datorat hemoragiei intraperitoneale după un traumatism abdominal închis. În cursul laparotomiei s-a descoperit ruptură splenică și un chist epitelial.

Concluzie: chisturile epiteliale splenice sunt entități rare și pot fi descoperite incidental în timpul tratamentului chirurgical al rupturii splenice și a hemoperitoneului. Aceste chisturi pot fi factori predispozanți ai rupturii splenice în caz de presiune intraabdominală crescută sau traumatisme abdominale închise.

Cuvinte cheie: chist epitelial splenic, ruptură splenică, descoperire incidentală, laparotomie

Abstract

Background/Aim: Primary splenic cysts are relatively uncommon and in the majority of cases are classified as epithelial cysts which are more frequent in young patients. Most of them are asymptomatic, and they are found incidentally during imaging techniques or laparotomy. We report two rare cases of patients with epithelial splenic cysts as an incidental finding during emergency laparotomy for splenic rupture.

Materials and Method: The first patient was a 70-year-old man who presented with symptoms of acute abdomen after intensive cough during an episode of asthma. Emergency laparotomy findings were splenic rupture and intraperitoneal haemorrhage. A splenectomy was performed and the histological examination showed a ruptured spleen with an epithelial cyst. The second patient was a 19-year-old man who was transferred to the Emergency Department in hypovolemic shock due to intraperitoneal bleeding after a blunt abdominal injury. During laparotomy a splenic rupture was found, and an epithelial cyst.

Conclusion: Epithelial splenic cysts are a rare entity and they can be an incidental finding during the surgical treatment of a spleen rupture and haemoperitoneum. These cysts may be

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a predisposing factor to splenic rupture in cases of increased intraabdominal pressure or blunt abdominal trauma.

Key words: epithelial splenic cyst, splenic rupture, incidental finding, laparotomy

Introduction

Splenic cysts are uncommonly encountered in the general surgical practice. Usually, splenic cysts are asymptomatic and can be found incidentally during abdominal ultrasonography, computed tomography or other imaging techniques. The symptoms are related to the size of the cyst and occasionally may be presented with complications such as infection, rupture and/or haemorrhage. In case of a large splenic cyst, the patient may complain about left upper quadrant discomfort, pain or tenderness. Sometimes large cysts may lead to splenomegaly, which may cause adjacent viscera displacement and pressure, leading to a variety of symptoms, including dyspnoea, shoulder pain and constipation. The most serious complication is splenic rupture, spontaneous or traumatic, with consequent intraabdominal bleeding that can be life-threatening. Laparotomy and splenectomy has been the treatment of choice for this serious complication. The incidental finding of a ruptured spleen with an epithelial cyst is a rare condition and there are no similar sources in the literature. We report two rare cases of patients with an epithelial splenic cyst as an incidental finding during emergency laparotomy for life threatening splenic rupture.

Presentation cases

Case 1

A 70-year-old man with a history of COPD arrived at the Emergency Department (ED) complaining of diffuse abdominal discomfort during the last month. He mentioned severe abdominal pain of increased intensity during the last two hours after prolonged cough. Physical examination showed generalized abdominal tenderness, without signs of abdominal injury. Bowel sounds were decreased. The patient was tachycardic and complained of pain in the left shoulder (Kehr sign). Chest and abdominal X-rays showed a mild elevation of the left hemidiaphragm. Computed Tomography scanning of the abdomen showed a large spleen with intraparenchymal haemorrhage and a cystic lesion in the lower pole of the spleen (Fig. 1). Furthermore, presence of fluid in the peritoneal cavity around the liver and at Douglas's pouch was shown.

The patient underwent an exploratory laparotomy and splenectomy. The spleen at histologic examination had a weight of 165 grams and measured 12 x 9 x 3 cm. A cyst was found in the lower pole of the spleen with a diameter of 5 cm and was characterized as epithelial. Multiple parenchymal

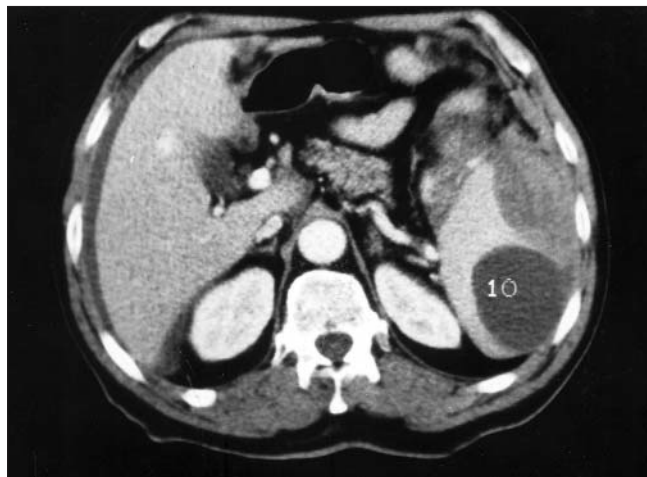


Figure 1. Patient 1. Abdominal Computed Tomography scanning of the abdomen shows a large spleen with intraparenchymal haemorrhage and a cystic lesion in the lower pole of the spleen

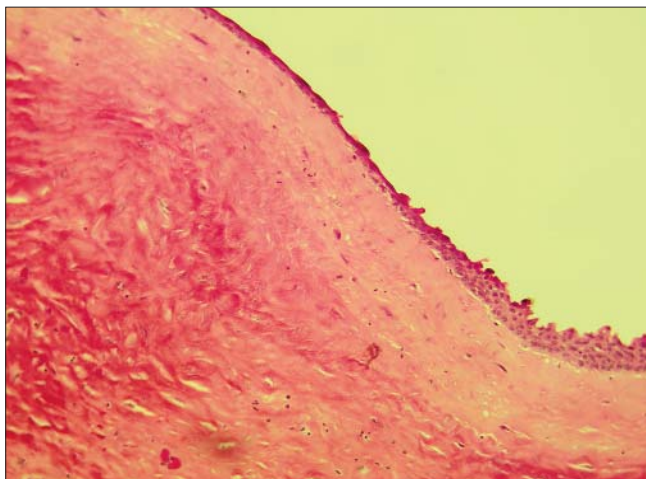


Figure 2. Patient 1. Histologic sections from other parts of the spleen show haemorrhagic infiltrations and sinusoid dilatations

ruptures were observed. Sections from other parts of the spleen showed haemorrhagic infiltrations and sinusoid dilatations (Fig. 2).

Case 2

A 19-year-old man was transferred to the ED after a motor vehicle accident and a blunt abdominal injury. Due to hemodynamic instability (BP: 85/50 mmHg and HR: 120 bpm) and a positive for haemorrhage diagnostic peritoneal lavage, the patient underwent emergency exploratory laparotomy. The intraoperative findings were a fragmented spleen (Fig. 3) containing a large whitish solid mass at the inferior pole (Fig. 3, 4). At pathologic examination the spleen contained an 11.5 x 6 cm cyst with a smooth outer surface and a hard white capsule 0.5 cm in thickness that covered the cyst almost in its entirety. The inner surface was smooth in

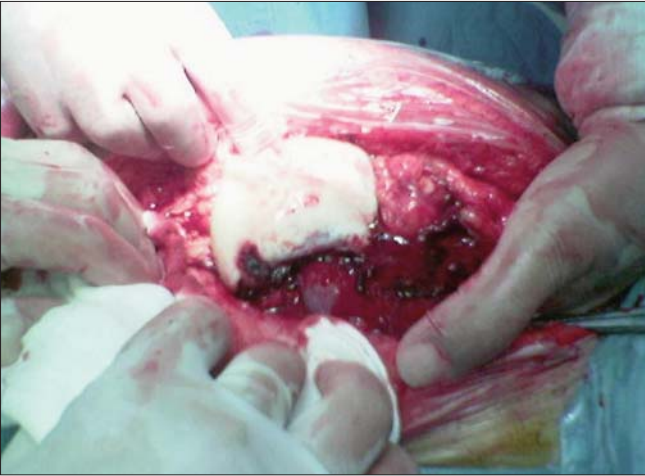


Figure 3. Patient 2. Intraoperatively, a fragmented spleen was found, containing a large whitish solid mass at the inferior pole

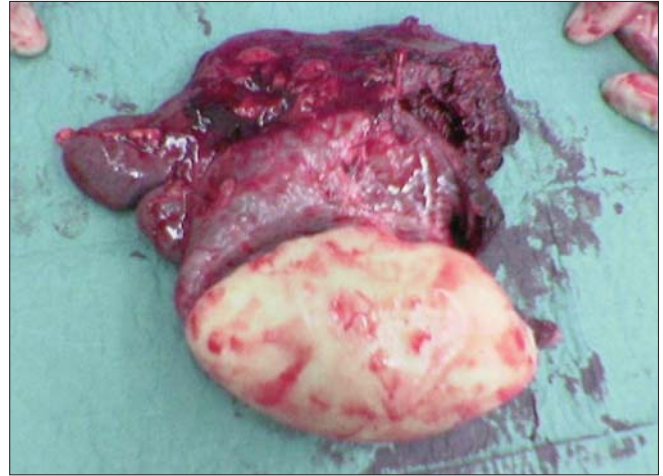


Figure 4. Patient 2. Spleen after resection, with the cyst in situ



Figure 5. Patient 2. Spleen after resection, with the cyst opened

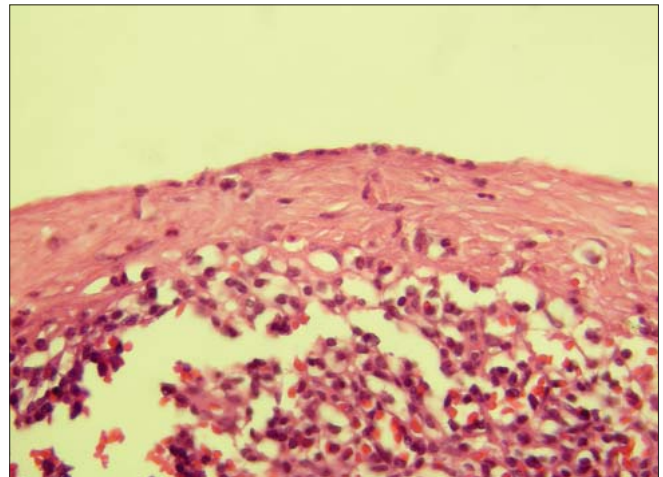


Figure 6. Patient 2. Histologic section of the cyst, with findings compatible with an epidermoid cyst

the area covered by the white capsule and the rest had an appearance similar to brain tissue (Fig. 5). Histology findings were compatible with an epidermoid cyst (Fig. 6).

Discussion

The Fowler splenic cysts and tumours classification (modified by Martin) divides them into two types: primary cysts (who are further subdivided into parasitic and non-parasitic) and secondary ones. The most common cause of splenic cysts globally are parasites - mainly *Echinococcus granulosus* (8). These cysts may be also classified according not only to aetiology, but also to the presence or absence of epithelium. They are thusly divided into true cysts or secondary cysts (pseudocysts). Pseudocysts comprise 75% of non parasitic splenic cysts. Their wall is made of a thick collagen layer with calcium sediments, without epithelial layering. Their content

is a mixture of blood and necrotic debris. They are solitary and asymptomatic in their majority. They may be posttraumatic (hemorrhagic or serous), degenerative (infarcts) or inflammatory (4, 8, 9).

True or main cysts are rare and are usually observed in children (6) or young adults with a higher frequency in males. From 800 splenectomies performed at Mayo Clinic in a period of 30 years only 4 cases of splenic cysts were found. They are usually solitary but sometimes are multiple. Cysts have been recorded even in accessory spleens. Microscopically, their wall comprises of cylindrical, cuboidal or squamous epithelium. Their histogenesis is unknown. It has been hypothesized that they are formed by inclusions during the embryologic origin of the spleen as a remnant of the myocardium of the right ventricle (7).

A splenic cyst may be totally asymptomatic and found incidentally during examination for other diseases.

Symptoms are usually atypical and related to the cyst's size and can be described as dull pain or a feeling of weight at the epigastrium and the left upper quadrant (2). Symptoms related to pressure on the stomach are rarer and comprise of anorexia, nausea, vomiting and loss of body weight. Occasionally a patient may present a splenic cyst complication such as inflammation, abscess formation, intracystic haemorrhage, or peritonitis secondary to cyst rupture. The most common inflammation is from bacteria of the Salmonella group (8). Splenic rupture, depending on its pathogenicity may be spontaneous due to increased intra-abdominal pressure as in our case #1 or traumatic as in our case #2.

The so-called spontaneous splenic cyst ruptures may occur even after minimal injury (cough, vomiting, sudden movements, and rigorous palpation) due to a capsular contusion, the capsule being already detached due to the hematoma.

This phenomenon appears mainly at diseases that may cause splenomegaly due to various reasons, such as congestive splenomegaly, splenic infarction, splenic carcinoma, amyloidosis, cyst, pseudocyst, mononucleosis, neoplasms such as leukaemia and lymphomas, myeloid metaplasia in myelofibrosis. During the past years it was quite common in malaria.

Splenic rupture causes intraperitoneal haemorrhage and if not promptly treated by total or subtotal splenectomy, it may lead to the patient's death. It is common in major injuries (motor vehicle accidents, falling from high altitude, etc) (1,5).

The diagnosis of splenic cysts can be ascertained by ultrasound examination, computed tomography or magnetic resonance imaging. In our first case, the diagnosis was made by CT scanning. In the second one the hemoperitoneum was identified by diagnostic peritoneal lavage and the final diagnosis of the splenic rupture with a cyst was made during laparotomy.

We observed that the cases of true splenic cysts are quite rare in the international literature (5). The use of methods

such as ultrasound and Computed tomography are essential for ascertaining diagnosis, although the wide use of ultrasound today has led to an increase in the incidence of splenic cysts by 1%.

The treatment of splenic cysts includes aspiration, drainage, excision of the cyst, and subtotal or total splenectomy (5). Small asymptomatic cysts found incidentally by imaging methods are not necessarily excised except if their wall is very thin and there is a danger of rupture. It is nonetheless recommended to excise them when they are larger than 5 cm (8), because in our opinion they predispose to a higher risk of rupture in condition of increased intraabdominal pressure. In symptomatic cysts or in imminent rupture, and of course when intrabdominal haemorrhage is present, total or subtotal splenectomy is mandatory.

References

1. Lam CM, Yuen ST, Yuen WK. Hemoperitoneum caused by spontaneous rupture of a true splenic cyst. *Hepatogastroenterology*. 1998;45(23):1884-6.
2. Klee FE, Osswald BR, Wysocki S. Splenic cyst--a classical "incidental finding". *Zentralbl Chir*. 1996;121(10):805-16. [Article in German]
3. Sirinek KR, Evans WE. Non parasitic splenic cysts - case report of epidermoid cyst with review of the literature. *Am J Surg*. 1973;126(1):8-13.
4. Bürrig KF. Epithelial (true) splenic cysts. Pathogenesis of the mesothelial and so-called epidermoid cyst of the spleen. *Am J Surg Pathol*. 1988;12(4):275-81.
5. Gidaro GS. Cystic splenic disease of surgical interest. *G Chir*. 1997;18(10):555-9. [Article in Italian]
6. Musy PA, Roche B, Belli D, Bugmann P, Nussle D, Le Coultré C. Splenic cysts in pediatric patients--a report on 8 cases and review of the literature. *Eur J Pediatr Surg*. 1992;2(3):137-40.
7. Walz MK, Metz KA, Eigler FW. Splenic cysts. Their morphology, diagnosis and therapy. *Dtsch Med Wochenschr*. 1991;116(37):1377-83. [Article in German]
8. Macheras A, Misiakos EP, Liakakos T, Mpistarakis D, Fotiadis C, Karatzas G. Non-parasitic splenic cysts: a report of three cases. *World J Gastroenterol*. 2005;11(43):6884-7.