

Primary non-parasitic splenic cyst: a case report

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Cystic disease of the spleen is a relatively rare disease. It is classified either as a true primary cyst or as a secondary pseudocyst. Most splenic cysts are pseudocysts, which have non-epithelial lining, and are caused by previous abdominal blunt trauma. Conversely, primary splenic cysts have epithelial lining and are subdivided into parasitic and non-parasitic cyst. Non-parasitic primary splenic cyst is considered congenital and comprises about 10% of all splenic cysts. Total or partial splenectomy is the treatment of choice, but parasitic infection must be excluded prior to an operation. In this present report, we described a symptomatic, large primary non-parasitic splenic cyst, which was surgically treated with partial splenectomy. (*Korean J Hepatobiliary Pancreat Surg 2013;17:139-141*)

Key Words: Spleen; Cyst

INTRODUCTION

Cystic disease of the spleen is relatively rare, with an incidence of 0.07%.¹ The splenic cysts categorized either as primary (true cysts) or as secondary (pseudocysts). The latter is mostly caused by abdominal trauma and has no epithelial lining in the cystic lumen. Conversely, a primary splenic cyst has epithelial lining of the lumen. Additionally, primary splenic cysts are further subdivided into parasitic and non-parasitic cyst. Non-parasitic primary splenic cyst is considered congenital and comprise of about 10% of all splenic cysts.² We present here a rare case of primary non-parasitic splenic cyst that is confirmed by pathologic diagnosis after partial splenectomy.

CASE

A 20-year-old female immigrant worker with a dull abdominal pain of several days duration was referred to our hospital from a local clinic, where an abdominal ultrasonography (US) had revealed a large cystic mass in the spleen. On abdominal examination, a soft non-tender mass was palpable in the left upper quadrant. Her medical history was notable only for appendectomy performed sev-

eral years ago. She had no other traumatic event in the abdomen. Her vital signs were stable and all laboratory findings were within the normal range. Abdominal computed tomography (CT) scan revealed the splenic cyst to measure 14 cm in diameter with scoliosis most likely due to mass effect (Fig. 1). The patient was offered open or laparoscopic partial resection of the spleen, and the patient opted for open surgery for economic reasons.

On laparotomy, the cyst was found to arise from the spleen. It was adhered to the left lateral side of the liver and left stomach wall (Fig. 2). The cystic fluid was carefully aspirated to avoid rupture into the operation field. After this, the whole cyst was excised in a partial splenectomy. A closed drain was left near the splenectomy site.

The result of serologic hydatid antibody test had been notified several days after the operation and was negative. Both carbohydrate antigen 19-9 (CA 19-9) and carcinoembryonic antigen (CEA) of the cystic fluid were over 1,000 U/ml respectively. The pathologic report indicated a primary splenic cyst with cystwalls containing stratified squamous epithelial cells (Fig. 3). The patient was discharged on the 10th postoperative day without complications.

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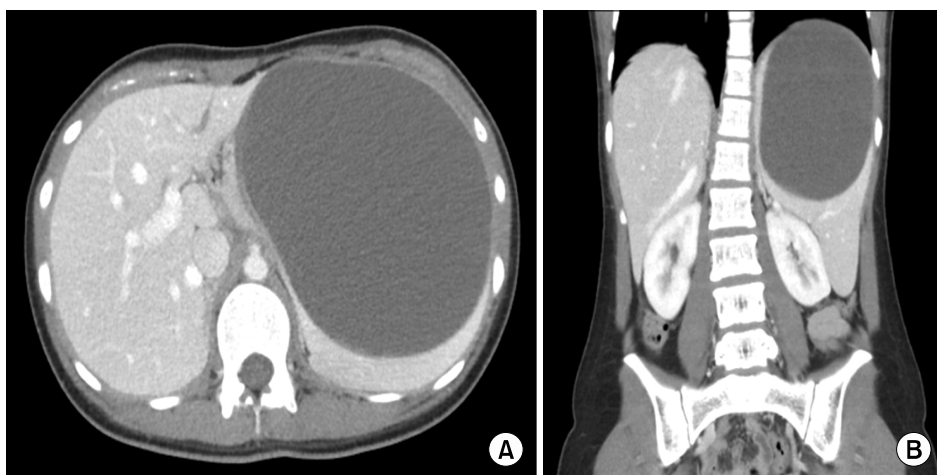


Fig. 1. Abdominal CT scan shows a large splenic cyst with scoliosis. (A) Axial view, (B) Coronal view.

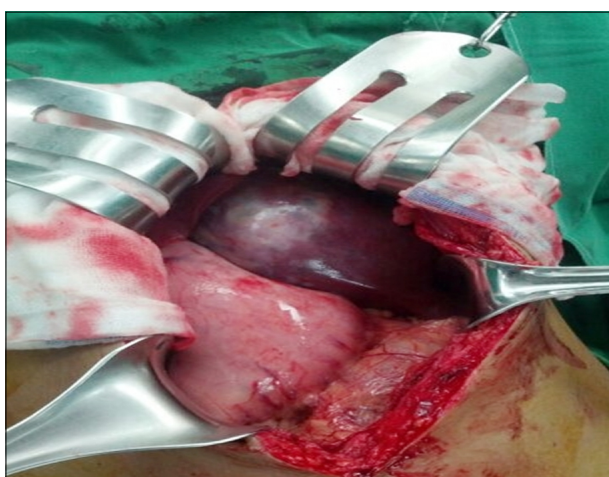


Fig. 2. The cyst was adhered to the left sides of the liver and stomach wall.

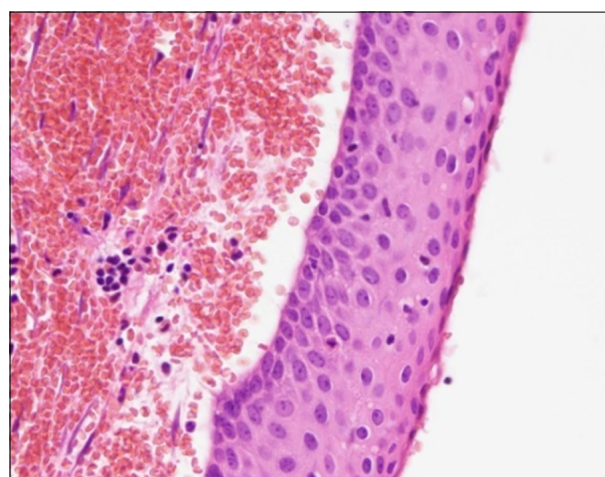


Fig. 3. The microscopic examination shows stratified squamous epithelial cells lining the cyst (H&E, $\times 400$).

DISCUSSION

Cystic diseases of the spleen are relatively rare with an incidence of 0.07%¹ and are categorized as either primary or secondary cysts.³ Secondary splenic cysts are usually results from previous abdominal trauma. These have no epithelial lining of the cystic lumen and, as such, are pseudocysts. Conversely, primary splenic cysts contain epithelial lining, and these true cysts are subdivided into parasitic and non-parasitic cyst.

Parasitic splenic cysts, or splenic hydatid disease (SHD), results from infection by *Echinococcus* species. It must be excluded before invasive procedure because spillage of the cystic contents may lead to anaphylactic shock or intraperitoneal dissemination of *Echinococcus* species.⁴ Although *Echinococcus* is not endemic to South Korea,

we had performed the serologic hydatid antibody test because the patient herself had come from an endemic country. However, the result was negative. During the operation, we took special care not to rupture the cystic fluid into the peritoneum, as the result of the serologic hydatid antibody test had not been made reported.

Primary non-parasitic splenic cysts are considered congenital and are lined by mesothelial, squamous, or transitional epithelium. The serum or cystic fluid is often positive for CA 19-9 and CEA, although these cysts do not have malignant potential.⁵⁻⁸ One hypothesis is that the inner epithelial cells produce these tumor markers, and these can circulate systematically by stoma-like channel between the lumen of the cyst and sinus of the adjacent splenic tissue.⁷ In our case, CA 19-9 and CEA levels from cystic fluid were high but pathologic report revealed no

malignant changes.

A large sized splenic cyst can be detected on physical examination. Moreover, preoperative US and CT scan are useful in understanding the size and relationship of the cystic lesions to surrounding structures.³ This information is helpful in the differential diagnosis and management. The conventional approach to splenic cysts has been complete total splenectomy. However, overwhelming post-splenectomy syndrome (OPSS) is a serious concern after total splenectomy, and partial splenectomy is increasingly considered as the treatment of choice.⁹ Additionally, laparoscopic approach has been performed successfully.^{6,9-11} Percutaneous drainage or injection of sclerosing agents have been used in limited cases for small cysts, but these procedures are associated with high recurrence rates.^{12,13}

In summary, splenic non-parasitic true cyst is a rare disease. It can be detected by ultrasonography or CT scan and are confirmed by pathologic findings. The serologic hydatid antibody is checked prior to an invasive procedure to exclude parasitic infection. Cystic tumor markers can be elevated in the serum, but it has no malignant potential. Partial or total splenectomy is the treatment of choice, and percutaneous drainage can be considered for small cysts or as bridges to surgery.

REFERENCES

1. Yoshikane H, Suzuki T, Yoshioka N, et al. Giant splenic cyst with high serum concentration of CA 19-9. Failure of treatment with percutaneous transcatheter drainage and injection of tetracycline. *Scand J Gastroenterol* 1996;31:524-526.
2. Shukla RM, Mukhopadhyay M, Mandal KC, et al. Giant congenital infected splenic cyst: An interesting case report and review of the literature. *Indian J Surg* 2010;72:260-262.
3. Hansen MB, Moller AC. Splenic cysts. *Surg Laparosc Endosc Percutan Tech* 2004;14:316-322.
4. Vezakis A, Dellaportas D, Polymeneas G, et al. Two cases of primary splenic hydatid cyst in Greece. *Korean J Parasitol* 2012;50:147-150.
5. Inokuma T, Minami S, Suga K, et al. Spontaneously Ruptured Giant Splenic Cyst with Elevated Serum Levels of CA 19-9, CA 125 and Carcinoembryonic Antigen. *Case Rep Gastroenterol* 2010;4:191-197.
6. Sardi A, Ojeda HF, King D Jr. Laparoscopic resection of a benign true cyst of the spleen with the harmonic scalpel producing high levels of CA 19-9 and carcinoembryonic antigen. *Am Surg* 1998;64:1149-1154.
7. Trompetas V, Panagopoulos E, Priovolou-Papaevangelou M, et al. Giant benign true cyst of the spleen with high serum level of CA 19-9. *Eur J Gastroenterol Hepatol* 2002;14:85-88.
8. Yigitbasi R, Karabicak I, Aydogan F, et al. Benign splenic epithelial cyst accompanied by elevated Ca 19-9 level: a case report. *Mt Sinai J Med* 2006;73:871-873.
9. Macheras A, Misiakos EP, Liakakos T, et al. Non-parasitic splenic cysts: a report of three cases. *World J Gastroenterol* 2005;11:6884-6887.
10. Iimuro Y, Okada T, Sueoka H, et al. Laparoscopic management of giant splenic true cyst with partial splenectomy: A case report. *Asian J Endosc Surg* 2013;6:226-230.
11. Kiriakopoulos A, Tsakayannis D, Papadopoulos S, et al. Laparoscopic management of a ruptured giant epidermoid splenic cyst. *JLSLS* 2005;9:349-351.
12. De Caluwé D, Phelan E, Puri P. Pure alcohol injection of a congenital splenic cyst: a valid alternative? *J Pediatr Surg* 2003;38:629-632.
13. Morandi E, Castoldi M, Merlini DA, et al. Is there a role of percutaneous drainage in non-parasitic splenic cysts? Case report. *G Chir* 2012;33:343-345.