

CASE REPORT

Laparoscopic Distal Pancreatectomy for Multiple Epithelial Cysts in an Intrapancreatic Accessory Spleen. A Case Report and Review of Literature

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ABSTRACT

Context Accessory spleen is a congenital abnormality consisting of normal splenic tissue in ectopic sites that is found in approximately 10-15% of the general population. However, an intrapancreatic accessory spleen has seldom been reported and multiple epithelial cysts in the intrapancreatic accessory spleen are extremely rare. **Case report** A 37-year-old woman with no clinical manifestations presented with two cystic lesions in the tail of the pancreas. The tumor markers CA 19-9 (251 U/mL) and SPAN-1 (38 U/mL) were increased. Computed tomography showed a multilocular cyst, 40 mm in size, and a unilocular cyst, 20 mm in size, in the tail of the pancreas and gallstones. The cystic component was hypointense on T1-weighted magnetic resonance images and hyperintense on T2-weighted magnetic resonance images. A laparoscopic distal pancreatectomy was performed with the presumptive diagnosis of a mucinous cystic neoplasm or an intraductal papillary mucinous neoplasm with gallstones. The pathological examination showed that the walls of the two cysts were covered with non-keratinized stratified squamous epithelium, surrounded by normal splenic tissue. The final pathological diagnosis was two epithelial cysts originating from an intrapancreatic accessory spleen. **Conclusions** Even though multiple masses were detected in the pancreatic tail, the possibility of epithelial cysts originating from an accessory spleen should be considered. Laparoscopic distal pancreatectomy might be a safe and effective procedure and provide good cosmetic result for a benign or low-grade malignant tumor in the pancreas.

INTRODUCTION

Accessory spleen is a congenital abnormality consisting of normal splenic tissue in ectopic sites that is found in approximately 10-15% of the general population. However, an intrapancreatic accessory spleen has seldom been reported and multiple epithelial cysts in the intrapancreatic accessory spleen are extremely rare. We herein report a case of 37-year-old woman with two epithelial cysts originating from an intrapancreatic accessory spleen who underwent laparoscopic distal pancreatectomy

CASE REPORT

An asymptomatic, 37-year-old woman was admitted to our hospital for surgery. In 2006, she was

determined to have two cystic lesions in the pancreatic tail, 15 and 13 mm in size, respectively. By 2012, these cystic lesions had grown to 40 and 20 mm in size, respectively. She had a history of mixed connective tissue disease and took 2 mg of prednisone per day. Blood chemistry and urinalysis were within normal limits. The tumor markers CA 19-9 (251 U/mL; reference range 0-37 U/mL) and SPAN-1 (38 U/mL; reference range 0-30 U/mL) were increased. Computed tomography showed a multilocular cyst, 40 mm in size, and a unilocular cyst, 20 mm in size, in the tail of the pancreas and gallstones. The cystic component was hypointense on T1-weighted magnetic resonance images and hyperintense on T2-weighted magnetic resonance images (Figure 1). On magnetic resonance cholangiopancreatography, these cysts did not connect with the main pancreatic duct and had no solid components. Fluorine-¹⁸fluorodeoxyglucose positron emission tomography (FDG-PET) showed no significant increase in the maximum standard uptake value. Suspecting a mucinous cystic neoplasm or an intraductal papillary mucinous neoplasm with gallstones, a laparoscopic distal

Received July 3rd, 2013 – Accepted August 20th, 2013

Key words Choristoma; Epidermal Cyst; Laparoscopy; Pancreatectomy; Spleen

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Figure 1. Magnetic resonance imaging showed a multilocular cyst, 40 mm in size, and a unilocular cyst, 20 mm in size, in the tail of the pancreas.

pancreatectomy was performed. First, the spleen and the pancreatic tail were mobilized from the retroperitoneum using the four-port technique laparoscopically (Figure 2). After the mobilization, the splenic artery was identified, double clipped, and ligated. The splenic vein was also clipped and dissected. After access was achieved through the 5 cm suprapubic incision, the pancreatic tail and spleen including the two cystic lesions were transected with a linear stapler (Endo GIA Ultra Universal Stapler, 30 mm Black, Covidien, Dublin, Ireland) (Figure 3) and extracted from the peritoneal cavity. A cholecystectomy was also performed. The operative time was 278 min and the blood loss was 50 mL.

Macroscopically, the cut surface of the tumor showed a unilocular cyst and a multilocular cyst completely surrounded with a brown solid component that resembled normal spleen. A histopathological examination showed that the walls of the two cysts were composed of fibrous tissue. The inner surface of the cyst was partly covered with non-keratinized stratified squamous epithelium, surrounded by normal splenic tissue.

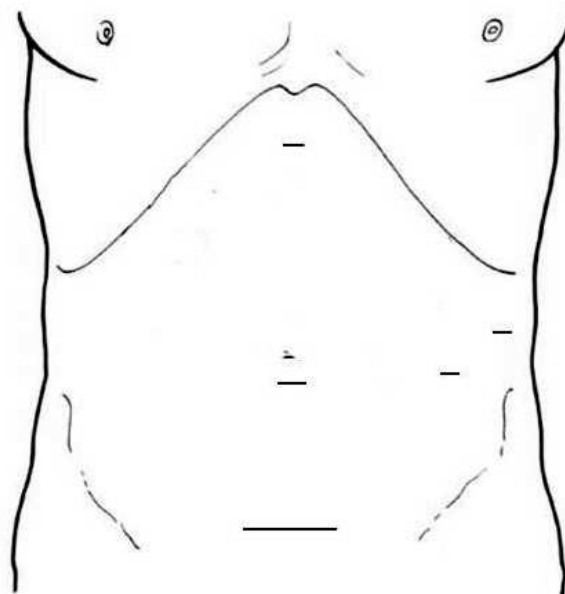


Figure 2. The laparoscopic procedure was performed using the four-port technique. The resected spleen and pancreatic tail were pulled out through a 5 cm long suprapubic incision.

The final pathological diagnosis was two epithelial cysts originating from an intrapancreatic accessory spleen (Figure 4). The patient had a pancreatic fistula (grade A, based on criteria of the International Study Group of Pancreatic Fistulas) and was managed conservatively. She was discharged on the 21st postoperative day. After the operation, her serum CA 19-9 levels decreased to normal. Over 12 months of follow-up, she continued to do well.

DISCUSSION

An accessory spleen is a congenital focus of healthy splenic tissue that is separated from the main body of the spleen. It results from failed fusion of the splenic anlage, which is located in the dorsal mesogastrium. This condition is observed in

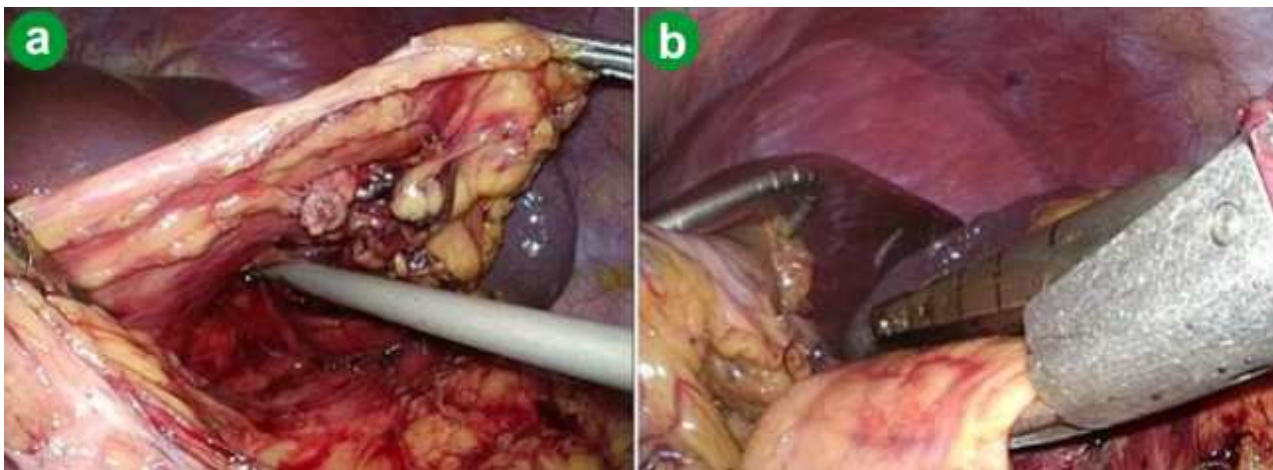


Figure 3. a. Laparoscopic view of the mobilization of the spleen and pancreatic tail. **b.** The pancreatic tail and spleen including the two cystic lesions have been transected with a linear stapler.

Table 1. Reported cases of an epithelial cyst or an epidermal cyst arising in an intrapancreatic accessory spleen.

| Case ID | Author, year | Age | Sex | Symptom | Size (cm) | Serum CA 19-9 | Form of cyst | Preoperative diagnosis | Surgery |
|---------|-----------------|-----|-----|---|-----------|---------------|--------------|---|---------|
| #1 | Davidson, 1980 | 40 | M | Nausea, vomiting, weight loss, anorexia | 7.0 | - | Multilocular | Pseudocyst, cystadenoma, cystadenocarcinoma | DP+S |
| #2 | Hamada, 1981 | 51 | M | Abdominal pain, vague | 6.0 | - | Unilocular | Pseudocyst | DP+S |
| #3 | Nakae, 1991 | 37 | F | Epigastric pain | 6.5 | - | Unilocular | Cystic mass | DP |
| #4 | Morohoshi, 1991 | 32 | F | Abdominal pain | 6.0 | Normal | Unilocular | Pancreatic tail cyst | CE |
| #5 | Tang, 1994 | 38 | M | Asymptomatic | 2.3 | - | Multilocular | Multiple cyst | DP+S |
| #6 | Higaki, 1998 | 46 | F | Left back pain | 3.0 | 201 | Multilocular | Pancreatic cyst | DP+S |
| #7 | Furukawa, 1998 | 45 | M | Asymptomatic | 2.0 | Normal | Multilocular | Malignant pancreatic tumor | DP |
| #8 | Tateyama, 1998 | 67 | F | Asymptomatic | 3.0 | Elevation | Multilocular | Malignant pancreatic tumor | DP+S |
| #9 | Sasou, 1999 | 49 | F | Asymptomatic | 4.3 | Normal | Multilocular | Cystic tumor | DP+S |
| #10 | Choi, 2000 | 54 | F | Epigastric pain, nausea, vomiting | 15.0 | - | | Benign cystic tumor | DP+S |
| #11 | Tsutsumi, 2000 | 51 | M | Asymptomatic | 2.5 | Normal | Multilocular | Cystic tumor, accessory spleen | DP+S |
| #12 | Horibe, 2001 | 48 | M | Asymptomatic | 2.0 | 53 | Unilocular | Mucin-producing tumor | DP+S+N |
| #13 | Fink, 2002 | 12 | F | Asymptomatic | 10 | - | Multilocular | Infected abdominal cyst | DP |
| #14 | Yokomizo, 2002 | 38 | M | Asymptomatic | 3.0 | 410 | Multilocular | Mucinous cyst adenoma, adenocarcinoma, epidermoid cyst | DP+S |
| #15 | Sonomura, 2002 | 45 | F | Epigastric pain | 3.5 | 159 | Multilocular | Cystadenocarcinoma, solid cystic tumor | DP+S |
| #16 | Kanazawa, 2004 | 58 | F | Asymptomatic | 2.5 | 62 | Multilocular | Mucinous cystic tumor | DP |
| #17 | Ru, 2007 | 41 | M | Asymptomatic | 2.5 | - | Unilocular | Well-circumscribed pancreatic tumor | DP+S |
| #18 | Servais, 2008 | 52 | F | Asymptomatic | 11.5 | - | Multilocular | Malignant pancreatic tumor | DP+S |
| #19 | Gleeson, 2008 | 32 | F | Right upper quadrant abdominal pain | 1.5 | - | Unilocular | Cystic pancreatic neoplasm | DP+S |
| #20 | Itano, 2008 | 40 | M | Asymptomatic | 4.0 | Normal | Unilocular | Epidermoid cyst in intrapancreatic accessory spleen | DP+S |
| #21 | Zhang, 2009 | 26 | F | Asymptomatic | 2.5 | Normal | Unilocular | Cystic lesion in the tail of the pancreas | DP |
| #22 | Itano, 2010 | 67 | M | Epigastric pain, weight loss | 1.5 | 182 | Unilocular | Epidermoid cyst in intrapancreatic accessory spleen | LADP+S |
| #23 | Kadota, 2010 | 57 | F | Asymptomatic | 6.0 | Normal | Multilocular | Pancreatic cystic tumor | DP+S |
| #24 | | 70 | F | Asymptomatic | 1.8 | 48 | Unilocular | Mucinous cystic tumor in the pancreas | DP+S |
| #25 | | 37 | M | Asymptomatic | 10.0 | 647 | Unilocular | Serous cystic tumor, lymphoepithelial cyst | DP |
| #26 | Horn, 2011 | 62 | M | Vague, left sided abdominal pain | 4.8 | - | Unilocular | Retroperitoneal mass with a possible cystic component | CE |
| #27 | Yamanishi, 2011 | 55 | F | Asymptomatic | 2.5 | 90 | Unilocular | Mucinous cystic neoplasm | DP+S |
| #28 | Iwasaki, 2011 | 36 | F | Asymptomatic | 3.4 | 79 | Multilocular | Mucinous cystic neoplasm | LDP |
| #29 | Urakami, 2011 | 50 | F | Asymptomatic | 3.0 | - | Unilocular | Epidermoid cyst, cystic tumor of in an intrapancreatic accessory spleen | LADP |
| #30 | Harris, 2012 | 39 | M | Asymptomatic | 2.5 | Normal | Unilocular | Cystic pancreatic lesion possibly malignant | LADP+S |
| #31 | Hu, 2012 | 25 | M | Asymptomatic | 4.2 | 1 | Unilocular | Cystic tumor | DP |
| #32 | | 43 | M | Asymptomatic | 5.2 | 18 | Unilocular | Cystadenoma | DP+S |
| #33 | | 59 | M | Asymptomatic | 4.6 | 78 | Multilocular | Cystic tumor | DP+S |
| #34 | | 34 | F | abdominal pain | 3.8 | 418 | Unilocular | Cystadenoma | DP+S |
| #35 | | 26 | F | Asymptomatic | 2.6 | 10 | Unilocular | Cystadenoma | DP |
| #36 | | 50 | M | Asymptomatic | 4.2 | 53 | Multilocular | Cystadenoma | DP+S |
| #37 | | 66 | M | Abdominal discomfort | 6.5 | 13 | Unilocular | Cystic tumor | DP+S |
| #38 | Hong, 2013 | 56 | F | Anemia | 4.0 | Normal | Unilocular | Pancreatic cancer | SPDP |
| #39 | Present case | 37 | F | Asymptomatic | 4.0 | 251 | Multilocular | Mucinous cystic neoplasm | LDP+S |
| | | | | | 2.0 | | Unilocular | Intraductal papillary mucinous neoplasm | |

CE: cyst excision; DP: distal pancreatectomy; F: female; LADP: laparoscopic-assisted distal pancreatectomy; LDP: laparoscopic distal pancreatectomy; PD: pancreaticoduodenectomy; M: male; N: nephrectomy; S: splenectomy; SPDP: spleen-preserving distal pancreatectomy

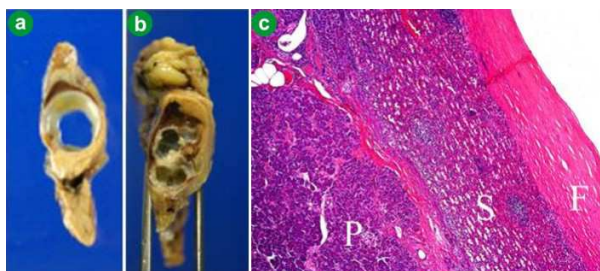


Figure 4. a. Unilocular cyst in the intrapancreatic accessory spleen. b. Multilocular cyst in the intrapancreatic accessory spleen. c. The histological section shows that the fibrotic wall (F) lined with non-keratinized stratified squamous epithelium is surrounded by accessory spleen tissue (S) in the pancreatic parenchyma (P).

10-30% of patients at autopsy. The splenic hilum is the most common site of an accessory spleen, followed by the pancreatic tail. In autopsy studies of 3,000 patients, 61 of 364 (17%) accessory spleens identified were found in the pancreatic tail [1]. Mortelé *et al.* performed abdominal CT scans in 1,000 consecutive patients. Of these patients, 156 (15.6%) had at least one accessory spleen, and intrapancreatic accessory spleens were seen in two patients (0.2%) [2].

However, cysts found within intrapancreatic accessory spleens are extremely rare. Nevertheless, a well-documented series of case reports can be found in the literature. Since the first case was described by Davidson *et al* [3] in 1980, 39 such cases, including the present report, have been documented in English language case reports (PubMed keywords: epithelial cyst, epidermoid cyst (epithelial inclusion cyst), intrapancreatic accessory spleen) [3, 4, 5, 6, 7, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24, 25, 26, 27, 28, 29, 30, 31, 32] (Table 1). However, to the best of our knowledge, this report is the first to describe

macroscopically multiple epithelial cysts within an accessory spleen.

A review of previously reported cases reveals that these lesions can occur equally in men and women (18 men, 21 women) and occur most frequently in middle age, with a median age of 45 years (range: 12 to 70 years). All of the lesions were located in the tail of the pancreas. Over half of the presentations were asymptomatic (26 of 39), and several patients had various symptoms including abdominal pain, epigastric pain or discomfort (n=10), weight loss (n=2), nausea and vomiting (n=2), and so on. Serum CA 19-9 levels were elevated in 15 of 39 cases. The median maximum diameter of the cystic lesions was 3.65 cm (range: 1.5 to 11.5 cm). This entity can mimic a cystic neoplasm of the pancreas, and it is often difficult to diagnose preoperatively. Only 3 of 39 cases had a precise diagnosis of epithelial cyst arising in an intrapancreatic accessory spleen. The remaining diagnoses were as follows: cystic tumor (n=11); mucinous cystic neoplasm, including a mucin-producing pancreatic tumor (n=7); cyst-adenoma/cystadenocarcinoma (n=6); benign/malignant pancreatic tumor (n=5); pancreatic cyst (n=4); or pancreatic pseudocyst (n=2).

The serum CA 19-9 levels were elevated preoperatively but decreased postoperatively in the present case, as well as in previous cases [8, 27]. Although further studies are needed to clarify the mechanism that elevates serum CA 19-9 levels, some cases might show this elevation, and this should be taken into consideration in the differential diagnosis.

Distal pancreatectomy was performed under a preoperative diagnosis of pancreatic cystic neoplasms, possibly malignant tumors, in most cases. Only five cases were managed laparo-

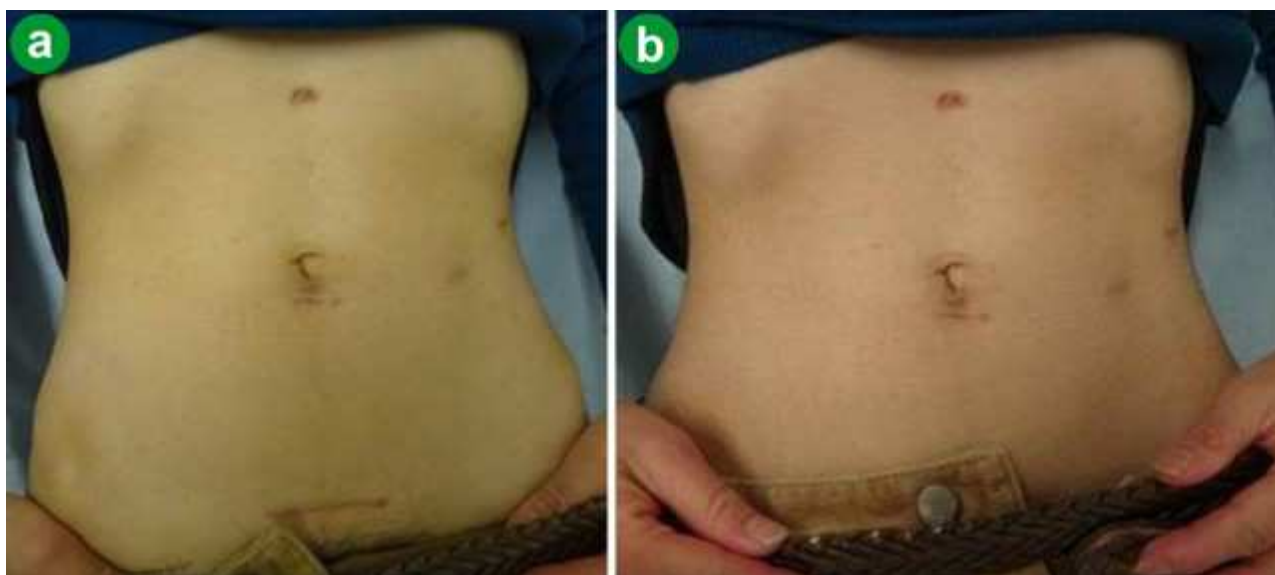


Figure 5. Postoperative scar at 3 months after surgery.

scopically [24, 28, 29, 30, 32]. Laparoscopic technique has made significant strides and is thought to an effective method of surgical management in cases of pancreatic and splenic cystic lesions, avoiding the shortcomings of open surgery [33]. For unspecified tumors, such as the current case, that appear benign, even though malignancy cannot be completely ruled out, a laparoscopic procedure would therefore be one of the best ways to avoid the drawbacks of open surgery, such as considerable pain, a prolonged hospitalization, and a poor cosmetic result. In the current case, the patient was a young woman and she benefited from the good cosmetic result of laparoscopic surgery (Figure 5).

In conclusion, when an asymptomatic intrapancreatic mass is detected, even if multiple masses are present, the possibility of epithelial cysts originating from an accessory spleen should be considered. Laparoscopic distal pancreatectomy might be a safe and effective procedure and provide a good cosmetic result for benign or low-grade malignant cysts in the pancreas.

Conflict of interest None of the authors have any conflicts of interest to declare

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