Laparoscopic Resection of an Epidermoid Cyst Arising from Intrapancreatic Accessary Spleen

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ABSTRACT

Context An epidermoid cyst arising from intrapancreatic accessory spleen is rare. Herein, we report the patient who had an epidermoid cyst arising from intrapancreatic accessory spleen with typical imaging features, which was diagnosed by preoperative imaging studies and successfully treated with minimally invasive surgery. **Case report** A seventy-five-year-old male was referred to our hospital presenting with left upper abdominal pain. Multidetector computed tomography highlighted a large cystic lesion with a diameter of 7 cm in the tail of the pancreas. Although the cystic components were low density and remained unenhanced during multiphase scans, a contrast enhancement pattern of the cystic walls was similar to that of the spleen on each phase. This patient was diagnosed with epidermoid cyst arising from intrapancreatic accessory spleen and underwent laparoscopic distal pancreatectomy and splenectomy to achieve pain-relief. The final pathological diagnosis was an epidermoid cyst arising from intrapancreatic accessory spleen. Postoperative course was uneventful, and his chief complaint completely resolved after surgery. **Conclusion** With recent advances of dynamic contrast imaging, the diagnosis of epidermoid cyst arising from intrapancreatic accessory spleen may be possible. In order to avoid unnecessary surgery, precise work-up and reliable preoperative diagnosis is necessary. When surgical resection is necessary to improve patient's symptoms, less invasive surgery with laparoscopic resection would be preferable.

INTRODUCTION

Accessory spleen is a congenital abnormality consisting of normal splenic tissue in ectopic sites [1]. In autopsy studies, it is seen in 11-16% of individuals and the tail of the pancreas is the second most common site of the accessory spleen [2, 3]. However, the development of an epidermoid cyst arising from intrapancreatic accessory spleen (ECIPAS) is extremely rare [4]. Herein, we report the patient who had an ECIPAS with typical imaging features, which was diagnosed by preoperative imaging studies and successfully treated with minimally invasive surgery.

CASE REPORT

A seventy-five-year-old male was referred to our hospital presenting with a one-month history of left upper abdominal pain. Laboratory data showed normal levels except carbohydrate antigen 19-9 increased to 39 U/mL (reference range 0-37 U/mL). Unenhanced and dual-phase-enhanced multidetector computed tomography (MDCT) scan was performed. The scan highlighted a large cystic lesion with a diameter of 7 cm in the tail of the pancreas **(Figure 1A-C)**. Stippled calcification was observed in the periphery. The cystic walls were thick and enhanced on each phase, while the cystic components were low density and remained unenhanced during multiphase scans. The cystic walls had higher density compared to the pancreatic parenchyma on the arterial phase **(Figure 1B)**. During the portal phase, the density of the cystic walls was comparable to the pancreatic parenchyma **(Figure 1C)**. The patterns of contrast enhancement of the cystic walls were similar to those of the spleen on each phase. T2-weighted magnetic resonance imaging (MRI) study revealed unilocular and high signal intensity in the cystic components **(Figure 2A)**. Diffusion-weighted imaging showed that restricted diffusion was observed in the cystic walls as well as in the spleen, but it was not evident in the cystic components **(Figure 2B)**. On the basis of these findings, this patient was diagnosed with ECIPAS and we recommended a routine follow up to him, while he wanted to achieve pain-relief by surgical intervention. He underwent laparoscopic distal pancreatectomy and splenectomy. Spleen preserving technique was not planned since preoperative MDCT scan revealed a large cystic lesion with a diameter of 7 cm extensively attached to the splenic artery and vein and the cyst was located close to the hilum of the spleen.

The patient was placed in the supine position and rotated 30° left side up with the table in a 30° head-up tilt. A five-trocar approach was used. First, the greater omentum

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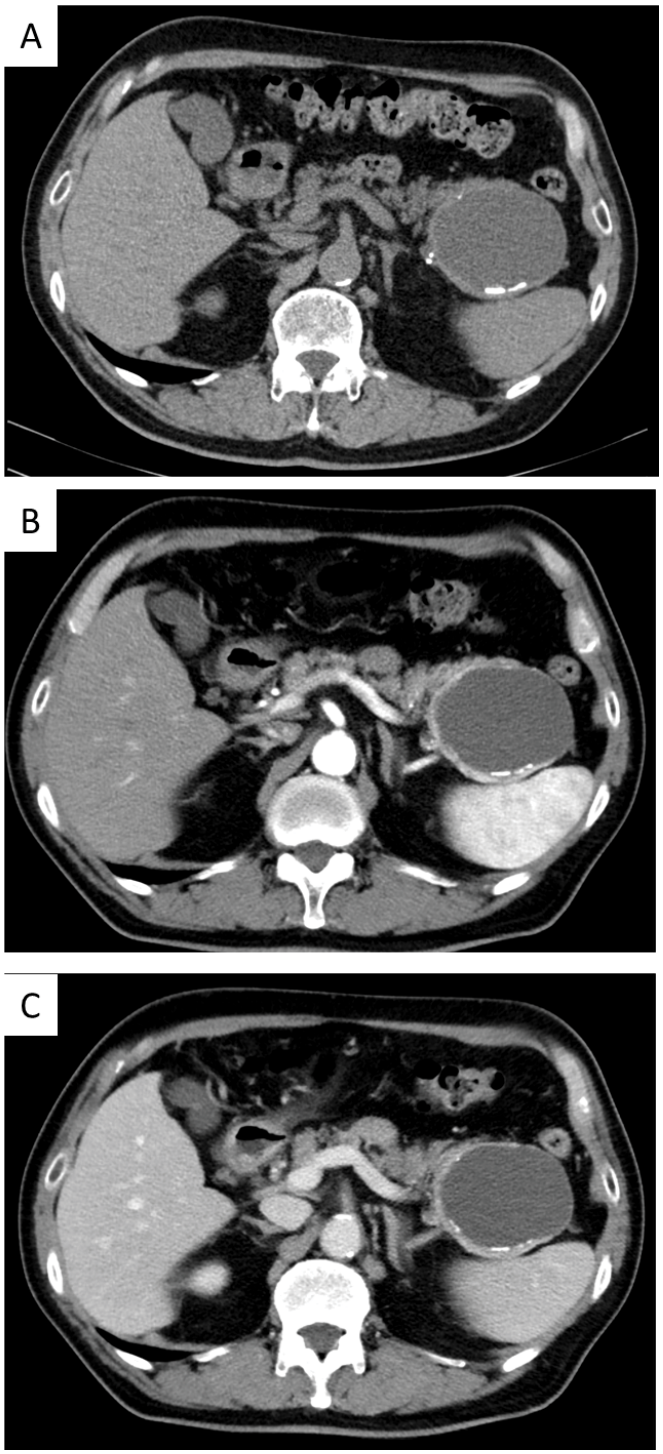


Figure 1. Preoperative multidetector computed tomography: **(a.).** Unenhanced scan, and **(b.).** arterial and **(c.).** portal phase.

was divided with laparoscopic coagulating shears. The left gastroepiploic artery was divided at its root after double clipping. All short gastric vessels were also divided and the stomach was completely retracted toward the cranial side, allowing the anterior aspect of the pancreas and a large cystic lesion located close to the splenic hilum to be exposed. Then, the lower margin of the pancreas was dissected from the medial to the left side, and splenocolic ligament was also divided. A dense adhesion was noted between epidermoid cyst and Gerota's fascia, suggesting the presence of persistent chronic inflammation **(Figure 3)**. Posterior dissection of the pancreas was continued to the cranial side to mobilize the distal part of the pancreas from the retroperitoneum. The splenic artery was identified at the upper border of the pancreas and excised after double clipping at the proximal side of the resection line. The pancreatic parenchyma was compressed and transected using the stapler (Endo-GIA Tri-Staple, black cartridge; Covidien, Norwalk, CT). After the distal pancreas and spleen were separated from the retroperitoneum, the whole specimens were removed. A closed-suction drain was placed at the site of the pancreatic stump. The duration of operation was 264 min and the amount of blood loss was 50 mL.

In the gross appearance of resected specimen after formalin fixation, cystic lesion was situated in the tail of the pancreas **(Figure 4A)**. Microscopic investigation revealed epidermoid cysts within accessory splenic tissue. The cystic walls were lined with mono- or multilayered epithelium comprised of cuboidal and squamous epithelium **(Figure 4B)**. They were surrounded by accessary splenic tissue in the pancreas parenchyma. No atypical or malignant changes were observed. The final pathological diagnosis was an ECIPAS.

Postoperative course was uneventful. Eventually, his chief complaint completely resolved after surgery.

DISCUSSION

An ECIPAS is a rare benign tumor and only 48 patients have been reported in the English literature to date [4-7]. In some of those patients, serum CA 19-9 level was

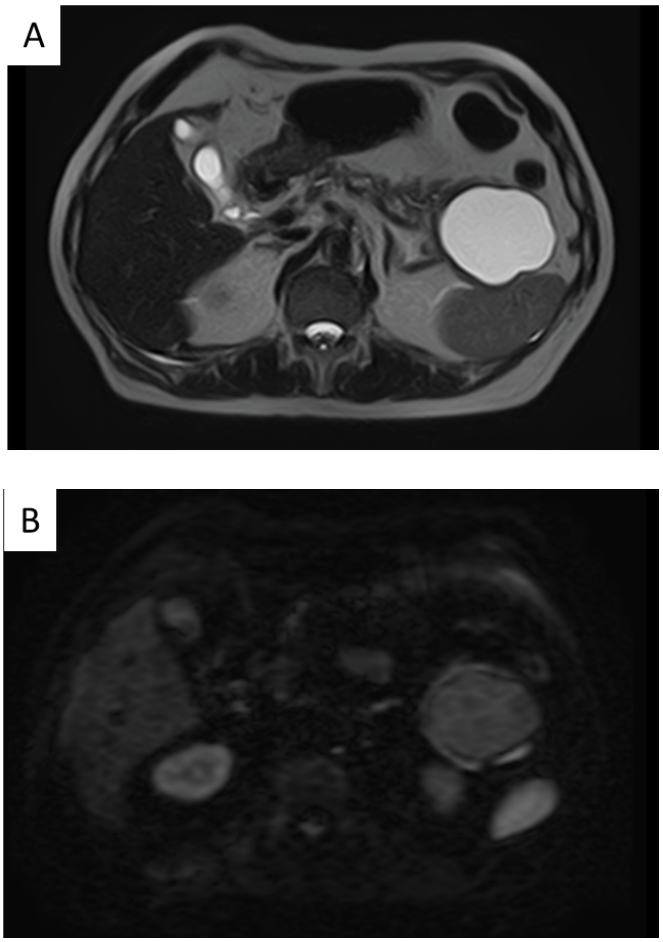
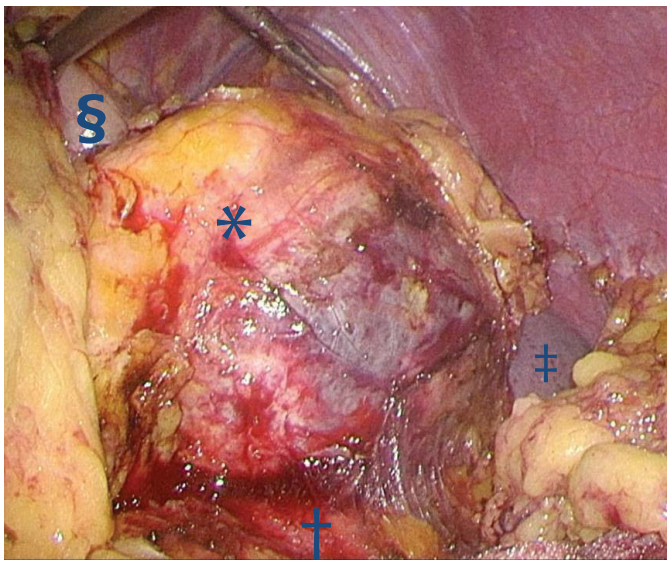


Figure 2. Preoperative magnetic resonance imaging. **(a.).** T2-weighted imaging and **(b.).** diffusion-weighted imaging.



*, †, §, and ‡ indicate epidermoid cyst, Gerota's fascia, stomach, and spleen, respectively.

Figure 3. Intraoperative photography. The omental bursa was opened and the stomach was retracted to the cranial side. The epidermoid cyst was dissected from the retroperitoneum.

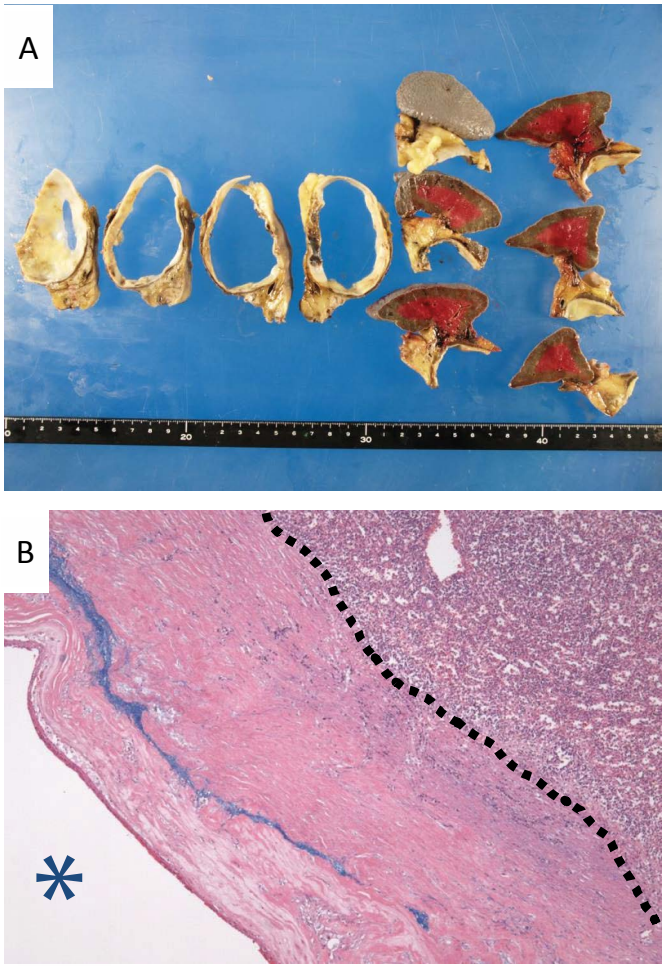


Figure 4: The macro- and microscopic appearances of surgical specimens: **(a.).** The macroscopic appearance of surgical specimens after formalin fixation shows cystic lesion situated in the tail of the pancreas. **(b.).** Microscopic appearance of the intrapancreatic cyst with Victoria blue-hematoxylin and eosin stain (magnification; x4). The area enclosed by dotted line indicates intrapancreatic splenic tissue.

* indicates the cystic cavity.

increased, which might be confused with malignancy. Indeed, approximately one third of patients with an ECIPAS who underwent pancreatectomy were preoperatively diagnosed as having pancreatic malignant neoplasm [4-7]. The differential diagnosis is cystic neoplasms, including intraductal papillary mucinous neoplasms, serous cyst neoplasms, and mucinous cyst neoplasms. Intraductal papillary mucinous neoplasms can be distinguished from ECIPS by magnetic resonance cholangiopancreatography since intraductal papillary mucinous neoplasms are normally connected to the pancreatic duct. Calcification is usually observed in serous cyst neoplasms and ECIPAS alike, but it is observed at the center of the cystic lesion in serous cyst neoplasms. Serous cyst neoplasms mostly contain multiple small cysts resulting in a lobulated contour and have the appearance of a central scar due to fibrous tissue retraction, which are different from ECIPAS [8]. Oligocystic serous cyst neoplasms, less common variant of serous cyst neoplasms, appear as larger and fewer cysts within the pancreas and it's imaging appearance may mimic that of an ECIPAS. However, the wall of any variants of serous cyst neoplasms are enhanced at delayed (portal) phase on contrast-enhanced CT/MR imaging [9]. The wall of ECIPAS is enhanced at early (arterial) phase since the cystic wall is completely equivalent to the splenic parenchyma. Mucinous cyst neoplasms commonly manifest as a unilocular or mildly septate cystic lesion. The wall of the cyst is typically thick and stippled calcification is sometimes observed. These radiological findings are similar to those of ECIPAS, and indeed, some of patients with ECIPAS were misdiagnosed as mucinous cyst neoplasms preoperatively and underwent surgery [4]. However, the enhancement pattern of the wall of mucinous cyst neoplasms is equivalent to that of serous cyst neoplasms,[9] which is different from ECIPAS. With currently-improved cross sectional techniques, better spatial resolution and dynamic contrast imaging, the diagnosis of ECIPAS may be possible when enhancing the cystic wall of the lesion similar to that of the spleen during multiphasic scans [5]. There has been no report concerning malignant change in ECIPS so far. In order to avoid unnecessary surgery, precise work-up and reliable preoperative diagnosis is necessary. Nevertheless, when surgical resection is necessary to improve patient's symptoms, less invasive surgery with laparoscopic resection would be preferable.

With recent advances of surgical techniques and endoscopic instruments, laparoscopic distal pancreatectomy has become widely accepted technique for the treatment of selected patients with pancreatic lesions [10-12]. Several meta-analyses compared between laparoscopic and open distal pancreatectomy have demonstrated the advantages of laparoscopic procedure. Venkat *et al.* performed meta-analysis of 18 studies including 1814 patients, 733(43%) of whom underwent laparoscopic and 1041 (57%) underwent open distal pancreatectomy [12]. The results suggested that patients undergoing laparoscopic distal pancreatectomy had lower blood loss during surgery and better postoperative recovery including reduced hospital length of stay and

time to oral intake. There was also lower risk of overall complications, surgical site infections, and incidence of readmission with the laparoscopic approach. More recently, Mehrabi *et al*. performed a meta-analysis using 29 retrospective studies including 2696 patients, 1328 of whom underwent laparoscopic and 1368 underwent open distal pancreatectomy [11]. In comparison with open surgery, laparoscopic surgery was a safe and effective approach in terms of operation time, blood loss, and perioperative mortality and morbidity, such as incidence of pancreatic fistula, fluid collection, postoperative bleeding, and wound infection. Laparoscopic surgery had also better postoperative recovery than open surgery with regard to the hospital length of stay and time to oral intake. In this context, Laparoscopic approach can be recommended as the treatment of choice for benign lesions in experienced surgeons when clinically appropriate. Indeed, laparoscopic approach may be superior to open approach in terms of good visibility of the operative field because of the magnifying effect and reduced venous blood loss owing to pneumoperitoneum pressure. Meanwhile, laparoscopic approach has some shortcomings including the lack of sensation and limitation of the two-dimensional field of view. Therefore, this approach is technically more demanding than conventional surgery and should be performed only by surgeons with sufficient experience in both open pancreatic surgery and advanced laparoscopic gastrointestinal surgery. Surgeons should not hesitate to convert a laparoscopic to an open procedure if patient safety is compromised. The rate of conversion to open surgery was approximately 14% (range 0–40%), mainly due to intraoperative massive bleeding [13-15].

Conflict of interest statement

Narita and other co-authors have no conflict of interest.

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