Laparoscopic spleen-preserving pancreatic resection for epidermoid cyst in an intrapancreatic accessory spleen: case report and literature review

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Introduction: An epidermoid cyst in an intrapancreatic accessory spleen (ECIPAS) is a rare non-neoplastic cyst, typically occurring in the pancreatic tail. It is difficult to preoperatively differentiate ECIPAS from other types of pancreatic neoplastic cysts.

Case presentation: We herein report a case of a 32-year-old man with a cystic tumor in the tail of the pancreas. The patient underwent a laparoscopic spleen-preserving distal pancreatectomy, and histological examination revealed the presence of ECIPAS. In addition, we also performed a literature review of 42 case reports of ECIPAS.

Conclusion: Although the preoperative diagnosis of ECIPAS is relatively difficult, familiarity with the imaging features, the clinical presentation and the location of the cyst could lead to a correct preoperative diagnosis of ECIPAS, which might thereby reduce the number of unnecessary resections.

Keywords: epidermoid cyst, accessory spleen, pancreas

Introduction
An epidermoid cyst in an intrapancreatic accessory spleen (ECIPAS) is extremely rare, with the prevalence of 1.7% in general population. The differential diagnosis of pancreatic cystic lesions is often challenging because of their similar findings on imaging. It is difficult to preoperatively differentiate ECIPAS from the “other” cystic neoplasms, such as a pancreatic pseudocyst, serous cystic neoplasm, mucinous cystic neoplasm, intraductal papillary mucinous neoplasm and lymphoepithelial cyst, or a solid pancreatic tumor, such as a pancreatic neuroendocrine tumor and solid pseudopapillary tumor, by using conventional imaging. Of the 51 cases of ECIPAS that have been reported in the English literature, only 5 cases were correctly diagnosed based on preoperative imaging. As ECIPAS is a non-neoplastic pancreatic cyst and has no malignant potential, a correct preoperative diagnosis could thereby reduce the number of unnecessary surgical resections of the pancreas. Herein, we report a case of a 32-year-old male with an ECIPAS and make a comprehensive review of the literature.

Case presentation
A 32-year-old male was admitted to The First Affiliated Hospital with a mass lesion on the pancreatic tail that was detected by abdominal ultrasound during an annual health checkup. No history of trauma or pancreatitis was recorded. He had normal vital signs and abdominal examination. Initial laboratory data also showed no
abnormalities, including those for tumor markers such as carcinoembryonic antigen (CEA) or carbohydrate antigen 19-9 (CA19-9). Abdominal computed tomography (CT) revealed a well-defined cystic neoplasm, which was located in the tail of pancreas and approaching to splenic hilum (Figure 1A), without enhancement in the arterial phase (Figure 1B) and the portal phase (Figure 1C). Additionally, endoscopic ultrasonography (EUS) showed a 3.5 cm multilocular cystic lesion in the pancreatic tail with an internal nodule (Figure 1D). The cystic tumor did not communicate with the main pancreatic duct.

Upon diagnosis of mucinous cystic neoplasms, the patient underwent a laparoscopic spleen-preserving distal pancreatectomy. Macroscopic analysis revealed that the mass in the tail of the pancreas was 4 cm at its greatest diameter and consisted of parenchymal and cystic components (Figure 2A). The gross pathology showed a well-demarcated, multilocular mass, containing colorless serous fluid. Microscopic analysis revealed a multilocular cyst surrounded by accessory splenic tissue in the pancreas parenchyma, and the cyst wall showed a thin multilayered squamous epithelium (Figure 2B). The final pathological diagnosis was epidermoid cyst originating from an intrapancræatic accessory spleen. His postoperative course was uneventful and he was discharged 8 days after the surgery.

**Ethical approval**

The study was approved by the ethics committee of The First Affiliated Hospital of Zhejiang University School of Medicine. Written informed consent was obtained from

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**Figure 1** The abdominal computed tomography (CT) scan confirmed a well-defined cystic neoplasm in the pancreatic tail (A), without enhancement in the arterial phase (B) and the portal phase (C). Endoscopic ultrasonography (EUS) showed a 3.5 cm multilocular cystic lesion in the pancreatic tail with an internal nodule (D).

**Figure 2** (A) Gross appearance of the epidermoid cyst in an intrapancræatic accessory spleen (ECIPAS), with 4 cm at its greatest diameter. (B) Microscopic analysis revealed a multilocular cyst surrounded by accessory splenic tissue in the pancreas parenchyma, and the cyst wall showed a thin multilayered squamous epithelium (H&E staining, ×50).
Discussion
Accessory spleens occur in ~10% of the population and can be found in various anatomic locations other than the splenic hilum. Approximately 20% of accessory spleens occur in or around the tail of the pancreas.1 Epidermoid cysts of the spleen are rare entities, comprising <10% of true non-parasitic splenic cysts.46 An ECIPAS is extremely rare, with only a few reports describing their clinical characteristics. Currently, with the advancement of imaging techniques, such as CT, magnetic resonance imaging (MRI) and EUS, an increasing number of ECIPASs have been detected.

Since Davidson et al2 reported the first case of ECIPAS in 1980, 41 articles and 50 patients have been reported in the English literature (Table 1). Including the present case, 20 cases were men and 32 cases were women. The mean age of the patients was 45.4 years (range 12–70 years), and 32 cases (61.5%) were younger than 50 years. Additionally, >50% of the cases were incidentally detected,6,7,10,12,13,16,17,19,22,24–26,29–31,33,35,39,41–43 while the other symptoms included nausea, vomiting, abdominal pain and discomfort, back pain, epigastric pain and fever. In all cases, the tumors were located on the pancreatic tail. The cyst appeared to be multilocular in 31 cases and unilocular in 14 cases (no information for 7 cases). The average cyst size was 4.3 cm (range 1.4–15 cm). As ECIPAS occurs at a relatively young age and it is located in the pancreatic tail, it is always necessary to differentiate ECIPAS when identifying a pancreatic tail cystic mass in young patients.

An elevation of serum CA19-9 level was observed in 21 cases,8,9,13,16–19,22,26,27,29,30,36,37,41,42 and hence, it was difficult to preoperatively differentiate between an ECIPAS and pancreatic malignancy during clinical analysis. Higaki et al8 reported that the serum CA19-9 levels markedly decreased to normal levels after surgery in patients diagnosed with an ECIPAS, a result suggesting that the serum CA19-9 might be secreted from the ECIPAS.

Most cases of ECIPAS are diagnosed after surgical resection based on the pathological characteristics. A preoperative imaging diagnosis of an ECIPAS is extremely difficult. Only 5 cases (9.8%) among the 51 reported cases were diagnosed preoperatively, while 1 out of 5 cases correctly diagnosed preoperatively was followed up without resection. Notably, in the present case, abdominal CT and EUS also revealed pancreatic mucinous neoplasm. As there are no characteristic features to define the lesion on radiology, it is difficult to entirely differentiate the cystic pancreatic malignancy prior to surgery and histopathological examination.

Until now, few studies have reported the imaging characteristics of ECIPAS. Hu et al47 noted that an accessory spleen surrounding the cyst was a key component for correct diagnosis, and therefore the relationship of enhancement between the splenic parenchyma and the parenchymal component of the lesion for the differential diagnosis of a cystic mass in the pancreatic tail was important. Itano et al21 described that 8 of 13 cases showed a solid tumor component upon CT or MRI, and several reports mentioned retrospectively that the images of the solid component were similar to those of the spleen. In our review, a diagnosis of a mucinous cystadenoma, cystadenocarcinoma, pseudocyst, neuroendocrine tumor or a potential malignant tumor was suspected in most cases.21 Interestingly, 3 out of 52 cases were diagnosed as ECIPAS preoperatively based on the similar density in the solid component and spleen on CT or MRI.21,37,43 Furthermore, Motosugi et al46 suggested that superparamagnetic iron oxide-based (SPIO) MRI was the most reliable tool for identifying an ECIPAS, because reticuloendothelial tissues including spleen took up SPIO and the signal intensity of the tissues changes (drops). Unfortunately, our patient did not receive MRI examination preoperatively. All the findings suggest that, in the presence of a relatively large amount of splenic tissues, a correct diagnosis would be possible based on a careful examination of images prior to surgery. However, relatively small amounts of splenic tissues may not be useful in the diagnosis. Therefore, ECIPAS should be considered in the differential diagnosis of pancreatic cystic lesions.

Until now, the treatment of ECIPAS consists of follow-up and surgical removal, including open or laparoscopic surgery with or without splenic preservation. No death has been reported during operation or in the short-term postoperative period. Fujii et al41 suggested that laparoscopic distal pancreatectomy could be a useful, minimally invasive surgical approach for treating pancreatic cysts as well as for the treatment of benign or low-grade malignant tumors located in the pancreatic body or tail. As ECIPAS was reported not to have malignant potential, a correct preoperative diagnosis could thereby avoid unnecessary surgery.

Conclusion
An ECIPAS is an extremely rare disease entity. Enhancing the cystic wall of ECIPAS similar to the spleen was a helpful feature. To prevent any unnecessary surgical intervention, it is important to recognize the ECIPAS as the differential diagnosis of pancreatic cyst. Familiarity with the imaging
| Case | Authors | Sex/Age | Symptom | Location | Size (cm) | Cyst | CA19-9 | CT           | MRI | Preoperative Diagnosis                  | Surgery |
|------|---------|---------|---------|----------|-----------|------|--------|--------------|-----|----------------------------------------|---------|
| 1    | Davidson et al | M/40    | Nausea  | Tail     | 5.5       | Multilocular | NI      | Cystic lesion surrounded by thin rim of tissue | NI   | Pseudocyst, cystadenoma and cystadenocarcinoma | DP      |
| 2    | Hanada et al | M/51    | Abdominal pain | Tail | 6       | NI | NI | Cystic mass with a rim of dense density | NI | Pseudocyst | DP      |
| 3    | Morohoshi et al | F/32    | Abdominal pain | Tail | 6       | Unilocular | Normal | Well-demarcated cystic lesion | NI | Pancreatic cyst | Cyst removal | SPDP |
| 4    | Nisk et al | F/37    | Epigastric pain | Tail | 6.5      | Unilocular | NI | Cystic lesion with a thin wall of high density | T1 low, T2 high | Pancreatic cyst | DP      |
| 5    | Tang et al | M/38    | Asymptomatic | Tail | 1.4      | Multilocular | NI | Well-demarcated hypodense lesion | NI | NI | DP      |
| 6    | Furukawa et al | M/45    | Asymptomatic | Tail | 2       | Multilocular | NI | Peripherally enhanced area, its density is equal to the spleen | NI | Primary cystic neoplasm | DP      |
| 7    | Higaki et al | F/46    | Left back pain | Tail | 3 | Multilocular | + | Oval nodule with a distinct margin | NI | Malignant tumor | DP      |
| 8    | Tateyama et al | F/67    | Abdominal pain | Tail | 3 | Multilocular | + | Cystic mass of low density | NI | NI | DP      |
| 9    | Sasou et al | F/49    | Asymptomatic | Tail | 4.3      | Multilocular | NI | NI | Pancreatic cyst tumor | DP      |
| 10   | Choi et al | F/54    | Epigastric pain | Tail | 15       | Multilocular | NI | Major cystic component, small solid component with the same homogeneous attenuation as in the spleen | Cyst: T1 low, T2 high; solid lesion: T1 low, T2 intermediate-high | Benign cyst of the pancreas or accessory spleen | DP      |
| 11   | Tsutsumi et al | M/51    | Asymptomatic | Tail | 2.5      | Multilocular | Normal | Well-demarcated cystic lesion containing a solid portion | Cystic lesion containing a solid portion | Benign cyst of the pancreas | DP      |
| 12   | Horibe et al | M/48    | Asymptomatic | Tail | 2 | Unilocular | + | No substance in the cyst by enhanced image | NI | Mucin-producing pancreatic tumor | DP      |
| 13   | Sonomura et al | F/45    | Epigastric pain | Tail | 3.5      | Multilocular | NI | Parenchymal medial lesion with calcification and cystic lateral lesion | NI | Cystadenocarcinoma or solid tumor of the pancreas | DP      |
| 14   | Fink et al | F/12    | Fever | Tail | 10       | Multilocular | NI | Rim enhancing cystic lesion, with a medial mural nodule | NI | Infected abdominal cyst | Cyst removal | SPDP |
| 15   | Yokomizo et al | M/38    | Asymptomatic | Tail | 3.0      | Multilocular | + | Cystic lesion containing a solid portion | MCN, adenocarcinoma and ECIPAS | MCN | SPDP |
| 16   | Kanazawa et al | F/58    | Asymptomatic | Tail | 2.5      | Multilocular | + | Septated low-density area | Cystic component: T1 hypo, T2 hyper | Mucinous cystadenoma and cystadenocarcinoma | DP      |
| 17   | Watanabe et al | F/55    | Postprandial epigastralgia | Tail | 3       | Multilocular | + | Multilocystic cystic tumor. No protruded lesion in the inner lumen | T1 low, T2 high | Mucinous cystadenoma and cystadenocarcinoma | DP      |
| No. | Authors | Gender | Age | Symptoms | Location | Size | Imaging Features | Diagnosis | Treatment |
|-----|---------|--------|-----|----------|----------|------|-----------------|-----------|-----------|
| 18  | Won et al. | M | 32 | Asymptomatic | Tail | 7.5 | Unilocular + | Well-circumscribed cystic mass with inner fluid debris or hemorrhagic fluid | NI | Pancreatic pseudocyst |
| 19  | Won et al. | F | 49 | Abdominal pain | Tail | 2.0 | Multilocular Normal | Well-circumscribed cystic tumor with septation | NI | Serous or mucinous cystadenoma |
| 20  | Ru et al. | M | 41 | Asymptomatic | Tail | 2.5 | Unilocular NI | Well-circumscribed tumor which partially compressed the spleen | NI | Cystic lesion of the pancreas |
| 21  | Itano et al. | M | 40 | Asymptomatic | Tail | 4.0 | Unilocular Normal | Solid component with the same homogeneous attenuation as the spleen | Cyst: T1/T2 high; solid component: T1 intermediate-low | ECIPAS |
| 22  | Servais et al. | F | 52 | Asymptomatic | Tail | 11.5 | Multilocular + | Gyriform mass which was thin walled and contained single peripheral septation | NI | Malignant pancreatic neoplasm |
| 23  | Gleeson et al. | F | 32 | Abdominal pain | Tail | 1.5 | Unilocular NI | Demarcated cyst without septation, calcification and satellite lesions | NI | Pancreatic cystic neoplasm |
| 24  | Zhang and Wang | F | 26 | Asymptomatic | Tail | 2.5 | Unilocular Normal | Cystic wall revealed a density similar to that of the pancreas | NI | Primary MCN |
| 25  | Reiss et al. | M | 49 | Asymptomatic | Tail | 3.6 | Multilocular NI | Heterogeneously enhancing mass | NI | MCN |
| 26  | Kadota et al. | F | 57 | Asymptomatic | Tail | 6 | Multilocular Normal | Cystic wall: a partial enhancement | NI | Pancreatic cystic tumor |
| 27  | Kadota et al. | F | 70 | Asymptomatic | Tail | 1.7 | NI + | Cystic mass lesion | NI | MCN |
| 28  | Kadota et al. | M | 37 | Asymptomatic | Tail | 10 | NI + | Cystic mass lesion with a partial enhancement of the cystic wall | NI | Serous cystic tumor or lymphoepithelial cyst |
| 29  | Itano et al. | M | 67 | Epigastric pain | Tail | 1.5 | Unilocular + | Cystic tissue and smooth solid component | Cyst: T1 intermediate, T2 high; Solid lesion: T1 intermediate-low | ECIPAS |
| 30  | Horn and Lele | M | 62 | Abdominal pain | Tail | 4.8 | Multilocular NI | Left-sided retroperitoneal mass with a possible cystic component | NI | MCN |
| 31  | Iwasaki et al. | F | 36 | Asymptomatic | Tail | 3.4 | Unilocular + | Septate low-density lesion, with an area showing higher degree of enhancement than the pancreas | NI | MCN |
| 32  | Yamanishi et al. | F | 55 | Asymptomatic | Tail | 2.5 | Unilocular + | Cyst wall was relatively thick, but not enhanced | Cyst: T1 slightly high, T2 strongly high | MCN |

(Continued)
| Case | Authors | Sex/age | Symptom | Location | Size (cm) | Cyst | CA19-9 | CT | MRI | Preoperative diagnosis | Surgery |
|------|---------|---------|---------|----------|-----------|------|--------|----|-----|-----------------------|---------|
| 33   | Urakami et al | F/50    | Asymptomatic | Tail     | 3.0       | Unilocular | NI    | Single cyst with a contrasted mass beside it | Cyst: T1 low, T2 high | ECIPAS | Laparoscopic SPDP |
| 34   | Khashab et al | F/49    | Abdominal pain | Tail     | 2.3       | Unilocular | NI    | Solid mass | NI | PNET | Laparoscopic SPDP |
| 35   | Harris et al | F/39    | Asymptomatic | Tail     | 2.5       | NI    | NI    | Stable hypodense lesion | Pancreatic cystic neoplasm | Malignant cystic tumor | Laparoscopic DP |
| 36   | Hong et al | F/54    | Abdominal discomfort | Tail     | 2         | Multilocular | NI    | Cystic mass | NI | NI | SPDP |
| 37   | Hamidian Jahromi et al | F/36  | Asymptomatic | Tail     | 5         | Multilocular | NI    | Cystic lesion | NI | NI | DP |
| 38   | Zavras et al | F/63    | Nausea and vomiting | Tail     | 12.6      | NI    | +     | Mass lesion with solid and cystic components | NI | Malignant tumor of the pancreas | SPDP |
| 39   | Kumamoto et al | M/39    | Diarrhea | Tail     | 3.8       | NI    | +     | A cyst lesion, surrounded by a crescent-like solid component with the same enhancement as the spleen | Typical findings of an intrapancreatic accessory spleen | ECIPAS | Laparoscopic SPDP |
| 40   | Kwak et al | F/21    | Abdominal pain and fever | Tail     | 2.5       | Multilocular | Normal | The wall of the cyst was relatively regular, thick and enhanced | Cyst: T1 iso, T2 hyper. Rim showed hyperintensity in DWI | SPT | Laparoscopic DP |
| 41   | Kato et al | F/33    | Asymptomatic | Tail     | 3         | Multilocular | Normal | The densities of the solid component and spleen on enhanced CT were similar | The intensity of the solid component on T1 and T2 was similar to that of the spleen | SPT and NET | Laparoscopic SPDP |
| 42   | Modi et al | F/62    | Abdominal pain | Tail     | 2.4       | NI    | NI    | Cystic lesion | NI | NI | Laparoscopic DP |
| 43   | Fujii et al | F/50    | Asymptomatic | Tail     | 4         | Unilocular | +     | A unilocular cystic lesion with same enhancement as the adjacent spleen | T1 low/T2 high | MCN | Laparoscopic SPDP |
| 44   | Fujii et al | F/60    | Back discomfort | Tail     | 3.5       | Multilocular | +     | A multilocular cystic lesion, solid component with enhancement similar to the spleen | Low T1 and high T2 | IPMN | Laparoscopic DP |
| 45   | Hirabayashi et al | M/38  | Asymptomatic | Tail     | 3         | Multilocular | Normal | NI | NI | NI | DP |
| 46   | Hirabayashi et al | F/40  | Abdominal pain | Tail     | 3.5       | Multilocular | +     | NI | NI | NI | Enucleation |
| 47   | Hirabayashi et al | F/39  | Asymptomatic | Tail     | 2         | Multilocular | +     | NI | NI | NI | DP |
features, the clinical presentation and the location of the
cyst will help radiologists make a more confident diagnosis.
Thus, making a definite preoperative diagnosis in most cases
is possible.

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