

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.¹ 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.²⁻⁴³ 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

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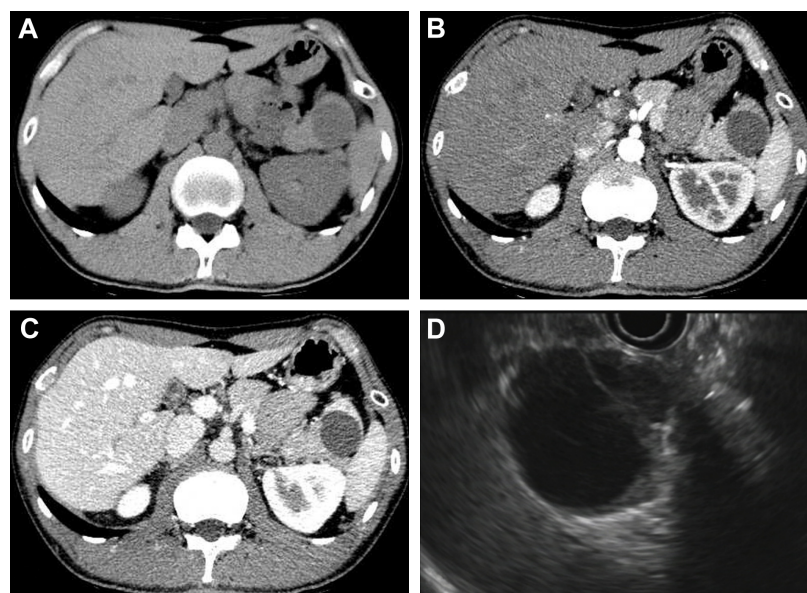


Figure 1 The abdominal computed tomography (CT) scan confirmed a well-defined cystic neoplasm in the pancreatic tail (**A**), without enhancement in the arterial phase (**B**) and the portal phase (**C**). Endoscopic ultrasonography (EUS) showed a 3.5 cm multilocular cystic lesion in the pancreatic tail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

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 intrapancreatic 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

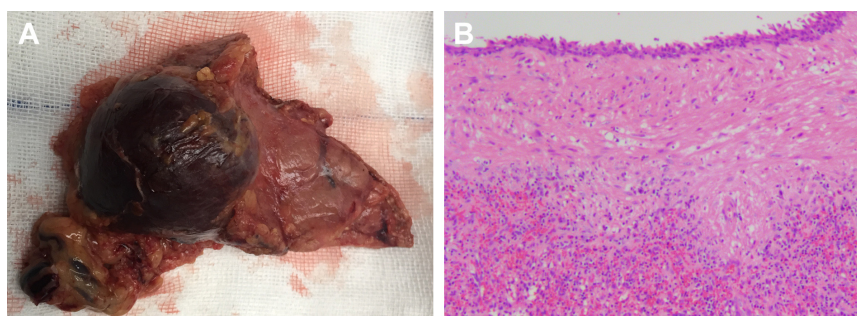


Figure 2 (**A**) Gross appearance of the epidermoid cyst in an intrapancreatic accessory spleen (ECIPAS), with 4 cm at its greatest diameter. (**B**) Microscopic analysis revealed a multilocular cyst surrounded by accessory splenic tissue in the pancreas parenchyma, and the cyst wall showed a thin multilayered squamous epithelium (H&E staining, ×50).

the patient to have the case details and any accompanying images published.

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.⁴⁴ 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 al⁴⁵ 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

Table 1 Reported studies of an ECIPAS in the English language literature

Case	Authors	Sex/ age	Symptom	Location	Size (cm)	Cyst	CA19-9	CT	MRI	Preoperative diagnosis	Surgery
1	Davidson et al ²	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 ³	M/51	Abdominal pain	Tail	6	NI	NI	Cystic mass with a rim of dense density	NI	Pseudocyst	DP
3	Morohoshi et al ⁴	F/32	Abdominal pain	Tail	6	Unilocular	Normal	Well-demarcated cystic lesion	NI	Pancreatic cyst	Cyst removal SPDP
4	Nakae et al ⁵	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 ⁶	M/38	Asymptomatic	Tail	1.4	Multilocular	NI	Well-demarcated hypodense lesion	NI	NI	DP
6	Furukawa et al ⁷	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 ⁸	F/46	Left back pain	Tail	3	Multilocular	+	Oval nodule with a distinct margin	NI	Malignant tumor	DP
8	Tateyama et al ⁹	F/67	Abdominal pain	Tail	3	Multilocular	+	Cystic mass of low density	NI	NI	DP
9	Sasou et al ¹⁰	F/49	Asymptomatic	Tail	4.3	Multilocular	NI	NI	NI	Pancreatic cystic tumor	DP
10	Choi et al ¹¹	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 ¹²	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 ¹³	M/48	Asymptomatic	Tail	2	Unilocular	+	No substance in the cyst by enhanced image	NI	Mucin-producing pancreatic tumor	DP
13	Sonomura et al ¹⁴	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 ¹⁵	F/12	Fever	Tail	10	Multilocular	NI	Rim enhancing cystic lesion, with a medial mural nodule	NI	Infected abdominal cyst	Cyst removal DP
15	Yokomizo et al ¹⁶	M/38	Asymptomatic	Tail	3.0	Multilocular	+	NI	Cyst: T2 super-high, cyst wall: delineated enhancement	MCN, adenocarcinoma and ECIPAS	DP
16	Kanazawa et al ¹⁷	F/58	Asymptomatic	Tail	2.5	Multilocular	+	Septated low-density area	Cystic component: T1 hypo, T2 hyper	MCN	SPDP
17	Watanabe et al ¹⁸	F/55	Postprandial epigastralgia	Tail	3	Multilocular	+	Multilocular cystic tumor. No protruded lesion in the inner lumen	T1 low, T2 high	Mucinous cystadenoma and cystadenocarcinoma	DP

18	Won et al ¹⁹	M/32	Asymptomatic	Tail	7.5	Unilocular	+	Well-circumscribed cystic mass with inner fluid debris or hemorrhagic fluid	NI	Pancreatic pseudocyst	SPDP
19	Won et al ¹⁹	F/49	Abdominal pain	Tail	2.0	Multilocular	Normal	Well-circumscribed cystic tumor with septation	NI	Serous or mucinous cystadenoma	Laparoscopic DP
20	Ru et al ²⁰	M/41	Asymptomatic	Tail	2.5	Unilocular	NI	Well-circumscribed tumor which partially compressed the spleen	NI	Cystic lesion of the pancreas	DP
21	Itano et al ²¹	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	DP
22	Servais et al ²²	F/52	Asymptomatic	Tail	11.5	Multilocular	+	Cystic mass which was thin walled and contained single peripheral septation	NI	Malignant pancreatic neoplasm	DP
23	Gleeson et al ²³	F/32	Abdominal pain	Tail	1.5	Unilocular	NI	Demarcated cyst without septation, calcification and satellite lesions	NI	Pancreatic cystic neoplasm	DP
24	Zhang and Wang ²⁴	F/26	Asymptomatic	Tail	2.5	Unilocular	Normal	Cystic wall revealed a density similar to that of the pancreas	NI	Primary MCN	SPDP
25	Reiss et al ²⁵	M/49	Asymptomatic	Tail	3.6	Multilocular	NI	Heterogeneously enhancing mass	NI	MCN	DP
26	Kadota et al ²⁶	F/57	Asymptomatic	Tail	6	Multilocular	Normal	Cystic wall: a partial enhancement	NI	Pancreatic cystic tumor	DP
27	Kadota et al ²⁶	F/70	Asymptomatic	Tail	1.7	NI	+	Cystic mass lesion	NI	MCN	DP
28	Kadota et al ²⁶	M/37	Asymptomatic	Tail	10	NI	+	Cystic mass lesion with a partial enhancement of the cystic wall	NI	Serous cystic tumor or lymphoepithelial cyst	DP
29	Itano et al ²⁷	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	Laparoscopic DP
30	Horn and Lele ²⁸	M/62	Abdominal pain	Tail	4.8	Multilocular	NI	Left-sided retroperitoneal mass with a possible cystic component	NI	NI	DP
31	Iwasaki et al ²⁹	F/36	Asymptomatic	Tail	3.4	Unilocular	+	Septate low-density lesion, with an area showing higher degree of enhancement than the pancreas	NI	MCN	Laparoscopic DP
32	Yamanishi et al ³⁰	F/55	Asymptomatic	Tail	2.5	Unilocular	+	Cyst wall was relatively thick, but not enhanced	Cyst: T1 slightly high, T2 strongly high	MCN	DP

(Continued)

Table 1 (Continued)

Case	Authors	Sex/ age	Symptom	Location	Size (cm)	Cyst	CA19-9	CT	MRI	Preoperative diagnosis	Surgery
33	Urakami et al ³¹	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 ³²	F/49	Abdominal pain	Tail	2.3	Unilocular	NI	Solid mass	NI	PNET	Laparoscopic SPDP
35	Harris et al ³³	F/39	Asymptomatic	Tail	2.5	NI	NI	Stable hypodense lesion	Pancreatic cystic neoplasm	Malignant cystic tumor	Laparoscopic DP
36	Hong et al ³⁴	F/54	Abdominal discomfort	Tail	2	Multilocular	NI	Cystic mass	NI	NI	SPDP
37	Hamidian Jahromi et al ³⁵	F/36	Asymptomatic	Tail	5	Multilocular	NI	Cystic lesion	NI	NI	DP
38	Zavras et al ³⁶	F/63	Nausea and vomiting	Tail	12.6	NI	+	Mass lesion with solid and cystic components	NI	Malignant tumor of the pancreas	DP
39	Kumamoto et al ³⁷	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 ³⁸	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 ³⁹	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 ⁴⁰	F/62	Abdominal pain	Tail	2.4	NI	NI	Cystic lesion	NI	NI	Laparoscopic DP
43	Fujii et al ⁴¹	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 ⁴¹	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 ⁴²	M/38	Asymptomatic	Tail	3	Multilocular	Normal	NI	NI	NI	DP
46	Hirabayashi et al ⁴²	F/40	Abdominal pain	Tail	3.5	Multilocular	+	NI	NI	NI	Enucleation
47	Hirabayashi et al ⁴²	F/39	Asymptomatic	Tail	2	Multilocular	+	NI	NI	NI	DP

48	Hirabayashi et al ⁴²	M/54	Asymptomatic	Tail	2.7	Multilocular	Normal	NI	NI	NI	Enucleation	
49	Hirabayashi et al ⁴²	M/55	Asymptomatic	Tail	3.5	Multilocular	+	NI	NI	NI	Enucleation	
50	Hirabayashi et al ⁴²	M/36	Asymptomatic	Tail	13.4	Multilocular	+	NI	NI	NI	DP	
51	Matsumoto et al ⁴³	F/40	Asymptomatic	Tail	1.5	Multilocular	Normal	A multilocular cystic lesion, solid periphery, with the same enhancement as the spleen		High T1- and T2-weighted images	ECIPAS	No
52	Our patient	M/32	Asymptomatic	Tail	3.5	Multilocular	Normal	A well-defined cystic neoplasm without enhancing mural nodes		NI	MCN	Laparoscopic SPDP

Note: +, higher than normal.

Abbreviations: CA19-9, carbohydrate antigen 19-9; CT, computed tomography; DP, distal pancreatectomy; DWI, diffusion weighted image; ECIPAS, epidermoid cyst in an intrapancreatic accessory spleen; F, female; IPMN, intraductal papillary mucinous neoplasm; M, male; MCN, mucinous cystic neoplasm; MRI, magnetic resonance imaging; NET, neuroendocrine tumor; NI, no information; PNET, pancreatic neuroendocrine tumor; SPDP, spleen preserving distal pancreatectomy; SPT, solid pseudopapillary tumor; hyper, hyperintensity; hypo, hypointensity.

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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Disclosure

The authors report no conflicts of interest in this work.

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