INTERNAL MEDICINE

□ CASE REPORT □

# Epidermoid Cyst in an Intrapancreatic Accessory Spleen: Case Report and Literature Review of the Preoperative Imaging Findings

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## Abstract

An epidermoid cyst arising within an intrapancreatic accessory spleen (ECIAS) is rare, and also difficult to correctly diagnose before surgery. It is mostly misdiagnosed as a cystic tumor, such as a mucinous cystic neoplasm or as a solid tumor with cystic degeneration, such as a neuro endocrine tumor. We herein report a case of ECIAS and also perform a literature review of 35 reports of ECIAS. Although the preoperative diagnosis of ECIAS using conventional imaging is relatively difficult to make, careful preoperative examinations of the features on computed tomography and magnetic resonance imaging could lead to a correct preoperative diagnosis of ECIAS which might thereby reduce the number of unnecessary resections.

**Key words:** intrapancreatic accessory spleen, epidermoid cyst, diagnostic imaging, imaging characteristics

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## Introduction

An epidermoid cyst arising within an intrapancreatic accessory spleen (ECIAS) is extremely rare. It is difficult to diagnose preoperatively using conventional imaging and thus is commonly misdiagnosed as an "other" cystic neoplasm, such as a mucinous cystic neoplasm (MCN), or a solid pancreatic tumor, such as a pancreatic neuroendocrine tumor (NET). Of the 38 cases (35 articles) of ECIAS that have been reported in the English literature, only 4 cases were correctively diagnosed based on preoperative imaging. Because ECIAS has no malignant potential, a correct preoperative diagnosis could thereby reduce the number of unnecessary surgical resections of the pancreas. We herein report a case of ECIAS that was preoperatively diagnosed as a neuroendocrine tumor or solid pseudopapillary neoplasm, and was resected using laparoscopic distal pancreatectomy. A literature review was also performed, focusing on the imaging characteristics of ECIAS that could be the key to making a correct preoperative diagnosis.

## Case Report

A 33-year-old, otherwise healthy, Japanese woman was referred to our hospital for further investigation of a mass lesion on the pancreatic tail that was detected by abdominal ultrasound during an annual health check. The patient had an unremarkable family history, including that of pancreatic neoplasms, and did not complain of any symptoms. The physical examination resulted in no abnormal findings. 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 ultrasound revealed a round-shaped mass lesion with a cystic component on the pancreatic tail. Contrast-enhanced computed tomography (CT) revealed a mass measuring approximately 3 cm in size in the pancreatic tail with a cystic

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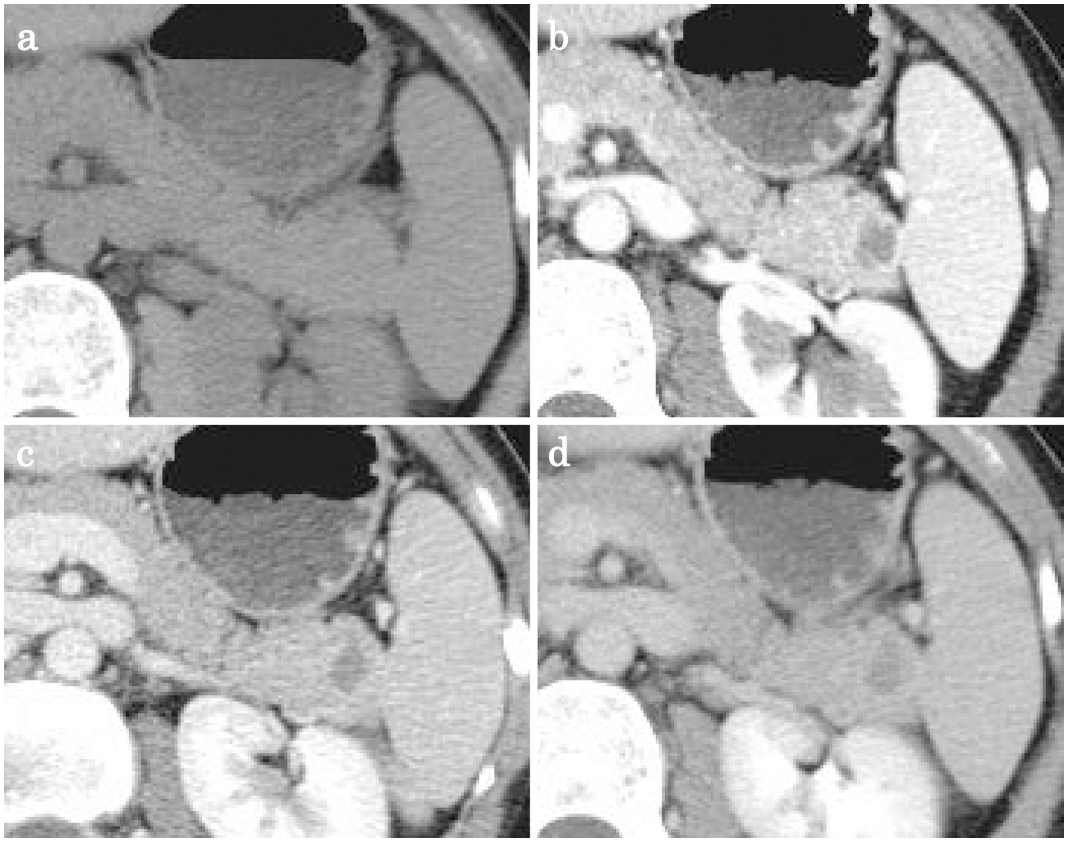


**Figure 1.** **Dynamic computed tomography reveal a mass measuring 3 cm in size in the pancreatic tail with a cystic lesion and a solid component located on the periphery that is enhanced in the arterial phase. The densities of the solid component and spleen are very similar (a: plain, b: arterial phase, c: portal phase, d: delayed phase).**

lesion and solid component located on the peripheral tumor that was enhanced in the early phase (Fig. 1). Magnetic resonance imaging (MRI) revealed that the cystic lesion was iso-intense on the T1-wighted image (WI) and hyper-intense on the T2-WI; the solid component was hypo-intense on T1-WI and slightly high on T2-WI (Fig. 2). On endoscopic ultrasonography (EUS), a round-shaped mass had a slightly high echoic solid component compared to the pancreas parenchyma, with a cystic lesion (Fig. 3a). EUS guided fine needle aspiration biopsy (EUS-FNA) was not performed, because it was difficult to puncture the mass while avoiding the rich perfusion of vessels around the mass lesion (Fig. 3b). As a result, the patient underwent laparoscopic distal pancreatectomy based on the diagnosis of solid peudopapillary neoplasm (SPN) or NET with cystic degeneration. The resected specimen revealed a well-demarcated 3 cm mass at its greatest diameter and a 1.5 cm multicystic lesion with brownish fluid (Fig. 4a). Microscopically, the solid component included splenic tissue with typical red and white pulp (Fig. 4b and c). The cyst was lined with a multilayered (2 to 5 layers) epithelium. The cyst wall was mainly composed of non-keratinized stratified squamous epithelium without any skin appendage (Fig. 4b and d), and the squamous epithelium was covered with a hobnail-like growth epithelium. No ovarian-type stroma was observed. In the cyst, blood, a cholesterin cleft and macrophages were observed; however, no hair was present. In an immunohistochemical (IH) analysis, the squamous epithelium of the cyst wall showed positive findings for CK5/6, p63 (Fig. 5), and negative findings for CK7, vimentin and muscle actin. The final pathological diagnosis was ECIAS, as no differentiation to the dermoid cyst and lymphoid tissue were observed.

## Discussion

An accessory spleen is a relatively common clinical presentation, found in almost 10% of the general population (1). Although most are observed in the splenic hilum, 17% of accessory spleens are located within the pancreatic tail (2). In contrast, an epidermoid cyst is a true cyst of the spleen. Typical histological findings are a unilocular or multilocular cyst lined with keratinized or non-keratinized stratified squamous epithelium surrounded by normal splenic tissue. The absence of hair and skin appendages in the cystic lesion and no lymphocyte infiltration are the key pathological features that differentiate an epidermoid cyst from a dermoid cyst and lymphoepitthelial cyst, respectively. ECIAS are extremely rare, with only a few reports describing their clinical characteristics. Since the first report of ECIAS was published by Davidson et al. in 1980 (3), 35 articles have been reported in the English literature (Table) (3-37). Including the present case, 15 cases were men and 24 cases were

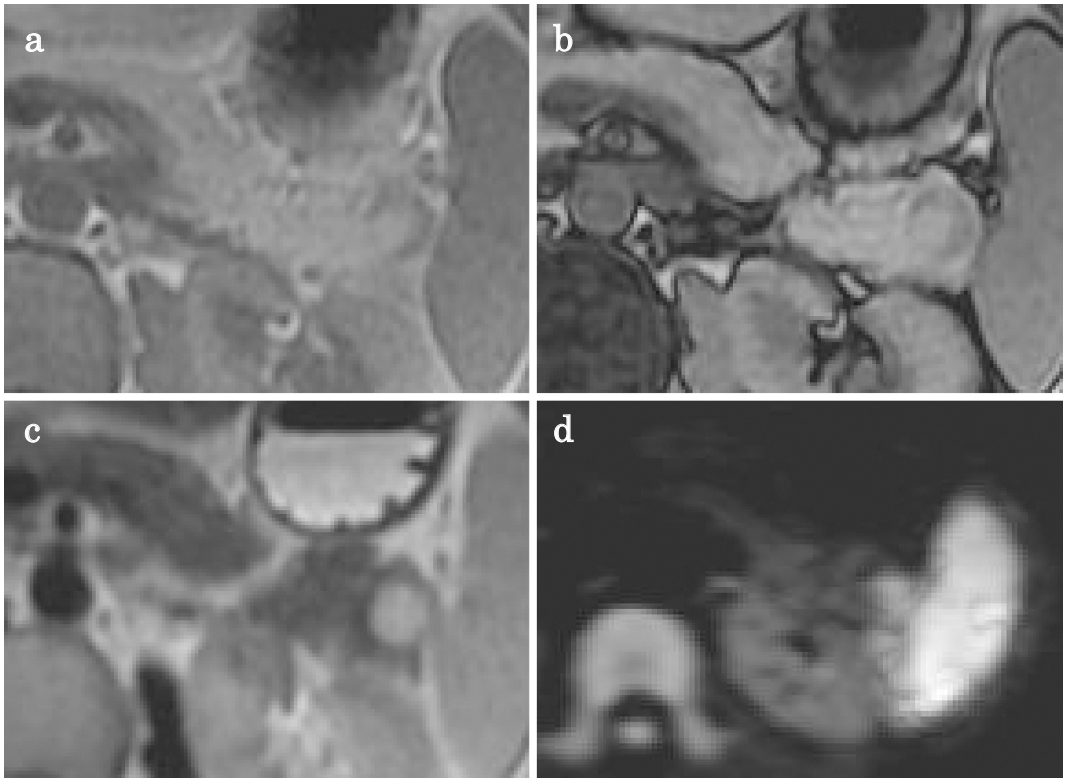


**Figure 2.** **Magnetic resonance images reveal that the intensity of the solid component on T1 weighted image and T2 weighted image is closely similar to that of the spleen and different from that of the pancreatic parenchyma (a: T1WI in phase, b: T1WI out of phase, c: T2WI, d: Diffusion WI).**

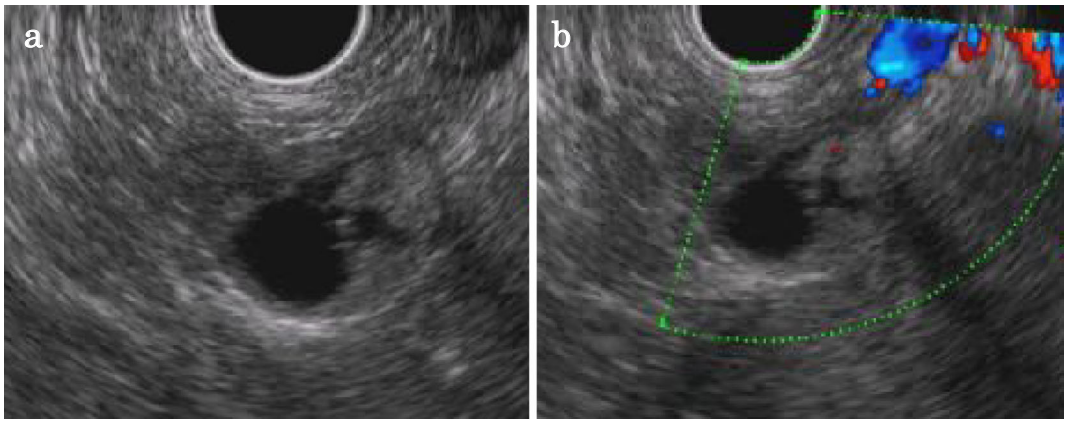


**Figure 3.** **Curved linear array endoscopic ultrasonography demonstrating a slightly high echoic component compared to the pancreas parenchyma, with a cystic lesion (a). It was difficult to puncture the mass while avoiding the rich perfusion of blood vessels around the mass lesion (b).**

women. The median age was 48 years, and 24 cases (61.5%) were younger than 50 years. In all cases, the tumors were located on the pancreatic tail. The cyst appeared to be multilocular in 21 cases and unilocular in 12 cases (no information for 6 cases). The average cyst size was 4.5 cm. Because ECIAS occurs at a relatively young age and it is located in the pancreatic tail, it is always necessary to differentiate ECIAS when identifying a pancreatic tail cystic mass in young patients.

Most cases of ECIAS are diagnosed after surgical resection based on the pathological characteristics. However, the correct preoperative diagnosis using conventional images such as CT and US is difficult in most cases. Only 4 cases (10.3%) among the 39 reported cases were correctly diagnosed using preoperative images.

Few studies have reported the imaging characteristics of ECIAS. Hu et al. analyzed the CT features of 7 consecutive patients with ECIAS; the cystic wall of the ECIAS showed a contrast enhancement similar to that of the spleen during multiphasic scans (38). In our review, 1 of 4 cases that were correctly, preoperatively diagnosed also had a similar density in the solid component and spleen on enhanced CT (23). In

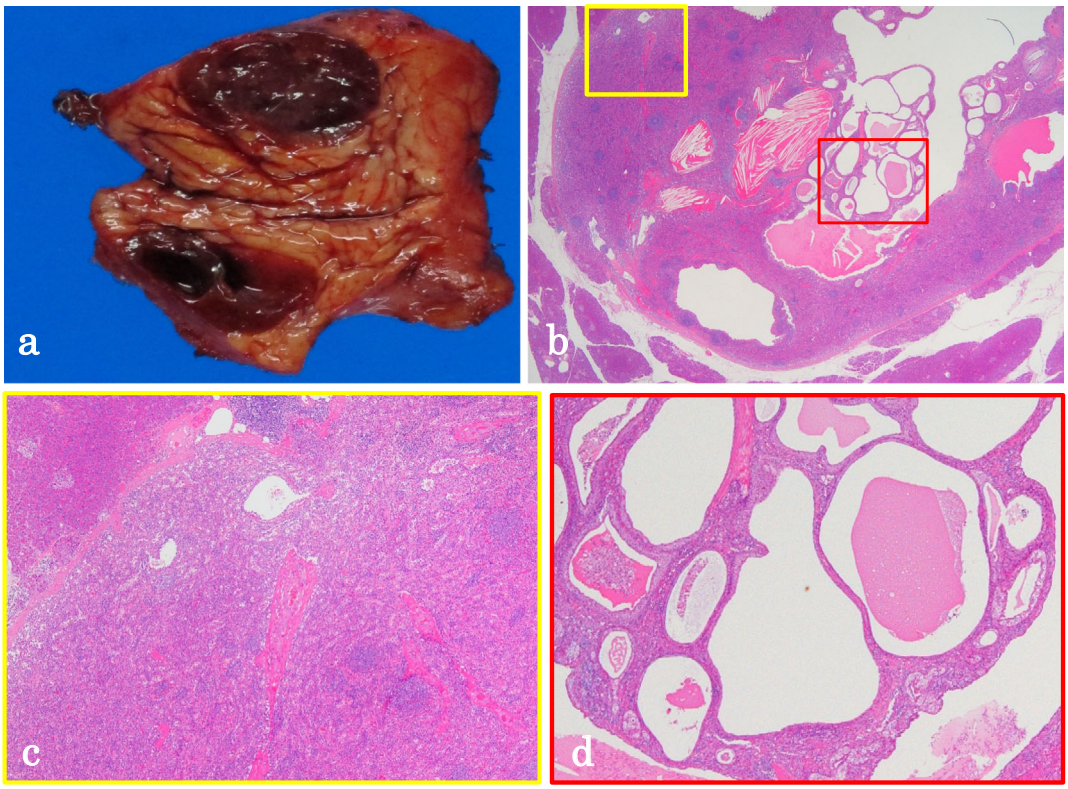


**Figure 4.** **The resected specimen reveals a well-demarcated 3 cm mass at its greatest diameter and a 1.5cm multicystic lesion with brownish fluid (a). The solid component includes splenic tissue with typical red and white pulp [b: Hematoxylin and Eosin (H&E) staining, ×10 magnification, c: H&E staining, ×40]. The cyst was multicystic and lined with a multilayered (two to five layers) epithelium (b: H&E staining, ×10, d: H&E staining, ×40).**

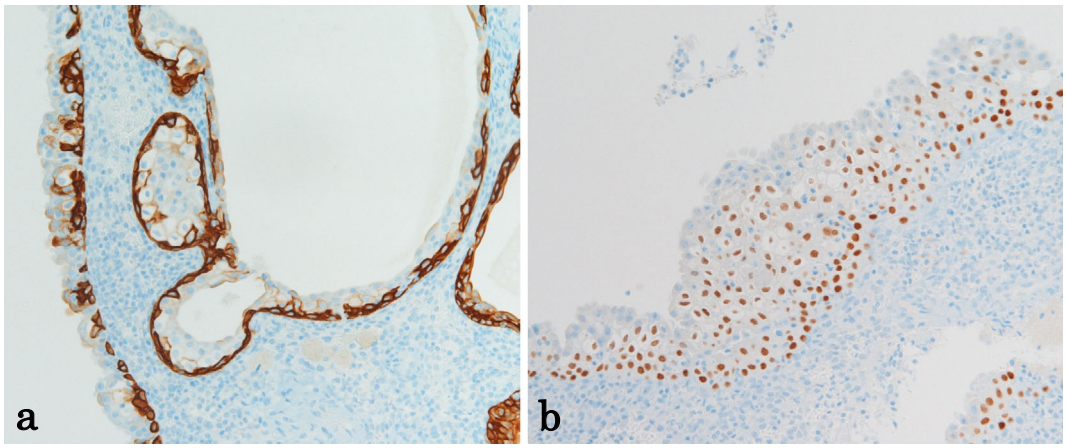


**Figure 5.** **The squamous epithelium of the cyst wall is positive for CK5/6 (a:×200) and p63 (b:×200).**

addition, Motosugi et al. described the MRI features of ECIAS, especially those of superparamagnetic iron oxide-based MRI; the solid component of the ECIAS showed the same intensity as that of the spleen (39). Based on similar MRI findings, a correct preoperative diagnosis was achieved for another case in our review (17). The similar density on enhanced CT and intensity on MRI between the solid component and the spleen might make it possible to make a correct, preoperative diagnosis of ECIAS.

The efficacy of EUS-FNA for the differential diagnosis of ECIAS has been investigated. Tatsas et al. reported 6 cases with a suspected intrapancreatic accessory spleen (IPAS) who underwent EUS-FNA (40). Of these 6 cases, IPAS was histologically confirmed for 3 cases. However, the FNA result of the case postoperatively diagnosed with ECIAS revealed only predominant macrophages and proteinaceous; therefore, no preoperative pathological or cytological evidence of ECIAS was obtained. In our review, EUS-FNA was performed for 4 cases (24-26, 32). However, a correct pathological diagnosis was not achieved in any of the cases.

**Table. Reported Literatures of an Epidermoid Cyst in an Intrapancreatic Accessory Spleen (36 Reports, 39 Cases).**

| Reference No. | Sex/Age | Symptom | Location | Size (cm) | Cyst | CT | MRI | Preoperative diagnosis | Surgery |
|---|---|---|---|---|---|---|---|---|---|
| 3 | M/40 | nausea | Tail | 5.5 | multilocular | cystic lesion surround by thin rim of tissue | NI | pseudocyst, cystadenoma, cystadenocarcinoma | DP |
| 4 | M/51 | abdominal pain | Tail | 6 | NI | well-defined cystic mass with a rim of dense density | NI | pseudocyst | DP |
| 5 | F/32 | abdominal pain | Tail | 6 | unilocular | expansively growing well-demarcated cystic lesion | NI | pancreatic cyst | cyst removal |
| 6 | F/37 | epigastric pain | Tail | 6.5 | unilocular | cystic lesion with a thin wall of high density | T1 low, T2 high | pancreatic cyst | SPDP |
| 7 | M/38 | NS | Tail | 1.4 | multilocular | well-demarcated hypodense lesion | NI | NI | DP |
| 8 | M/45 | NS | Tail | 2 | multilocular | peripherally enhanced area, its density is equal to the spleen | NI | primary cystic neoplasm | DP |
| 9 | F/46 | left back pain | Tail | 3 | multilocular | ovale nodulewith a distinct margin | NI | malignant tumor | DP |
| 10 | F/67 | abdominal pain | Tail | 3 | multilocular | cystic mass of low density | NI | NI | DP |
| 11 | F/49 | NI | Tail | 4.3 | multilocular | NI | NI | NI | DP |
| 12 | F/54 | epigastric pain | Tail | 15 | multilocular | small solid component with the same homogeneous attenuation in the spleen. | cyst:T1 low , T2 high, solid lesion:T1 low, T2 intermediate-high | benign cyst of the pancreas, or accessory spleen | DP |
| 13 | M/51 | NS | Tail | 2.5 | multilocular | well-demarcated cystic lesion containing a solid portion | cystic lesion containing a solid portion | benign cyst of the pancreas | DP |
| 14 | M/48 | NI | Tail | 2 | unilocular | reveal no substance in the cyst by enhanced image | NI | mucin-producing pancreatic tumor | DP |
| 15 | F/45 | epigastric pain | Tail | 3.5 | multilocular | parenchymal medial resion with calcification and cystic lateral resion | NI | cystadenocarcinoma, solid cystic tumor | DP |
| 16 | F/12 | fever (incidental) | Tail | 10 | multilocular | rim enhancing cystic lesion, with a medial mural nodule | NI | infected pseudocyst | cyst removal |
| 17 | M/38 | NI | Tail | 3 | multilocular | NI | cyst: T2 super-high, cyst wall: delineated enhancement. | MCN, adenocarcinoma, ECIAS | DP |
| 18 | F/58 | NS | Tail | 2.5 | multilocular | septated low density area | cystic component: T1 hypo, T2 hyper | MCN | SPDP |
| 19 | F/55 | epigastric pain | Tail | 3 | multilocular | multilocular cystic tumor. No protruted lesion in the inner lumen | cystic tumor: T1 low, T2 High | mucinous cystadenoma, adenocarcinoma | DP |
| 20 | M/32 | abdominal pain | Tail | 7.5 | unilocular | well circumscribed cystic mass with inner fluid debris or hemorrhagic fluid | NI | pseudocyst | SPDP |
| 20 | F/49 | abdominal pain | Tail | 2 | multilocular | well circumscribed cystic tumor with septation | NI | serous or mucinous cystadenoma | laparoscopic DP |
| 21 | M/41 | NS | Tail | 2.5 | unilocular | well-circumscribed tumor and partially compressed the spleen | NI | NI | DP |
| 22 | F/52 | NS | Tail | 11.5 | multilocular | cystic mass which was thin walled and contained single peripheral septation | NI | pancreatic malignancy | DP |
| 23 | M/40 | NS | Tail | 4 | unilocular | solid component that shows the same homogeneous attenuation as the spleen | cyst: T1 and T2 high solid component: T1 intermediate-low | ECIAS | DP |
| 24 | F/32 | abdominal pain | Tail | 1.5 | unilocular | demarcated cyst without septation, calcification, satelite lesions | NI | cystic pancreatic neoplasm | DP |
| 25 | F/26 | NS | Tail | 2.5 | unilocular | cystic wall revealed a density similar to that of the pancreas | NI | MCN | SPDP |

| Reference No. | Sex/Age | Symptom | Location | Size (cm) | Cyst | CT | MRI | Preoperative diagnosis | Surgery |
|---|---|---|---|---|---|---|---|---|---|
| 26 | M/49 | NS | Tail | 3.6 | multilocular | heterogeneously enhancing mass | NI | MCN | DP |
| 27 | F/57 | NS | Tail | 6 | multilocular | The cystic wall showed a partial enhancement | NI | pancreatic cystic tumor | DP |
| 27 | F/70 | NS | Tail | 1.7 | NI | cystic mass lesion | NI | MCN | DP |
| 27 | M/37 | NS | Tail | 10 | NI | cystic mass lesion with a partial enhancement of the cystic wall | NI | serous cystic neoplasm, lymphoepithelial cyst | DP |
| 28 | M/67 | epigastric pain | Tail | 1.5 | unilocular | cystic tissue and smooth solid component which was clearly seen in CECT | cyst: T1 intermediate, T2 high. Solid lesion: T1 intermediate-low | ECIAS | laparoscopic DP |
| 29 | M/62 | abdominal pain | Tail | 4.8 | multilocular | left sided retroperitoneal mass with a possible cystic component | NI | NI | DP |
| 30 | F/55 | NS | Tail | 2.5 | unilocular | cyst wall was reratively thick, but not enhanced | cyst: T1 slightly high, T2 strongly high | MCN | DP |
| 31 | F/36 | left hypo-chondralgia | Tail | 3.4 | unilocular | septate low-density lesion, with an area showing higher degree of enhancement than the pancreas | NI | MCN | laparoscopic DP |
| 32 | F/49 | abdominal pain | Tail | 2.3 | NI | solid mass | NI | PNET | laparoscopic SPDP |
| 33 | F/50 | NS | Tail | 3 | unilocular | single cyst with a contrasted mass beside it | cyst: T1 low, T2 high | ECIAS | laparoscopic SPDP |
| 34 | M/39 | NS | Tail | 2.5 | NI | stable hypodense lesion | pancreatic cystic neoplasm | malignant cystic tumor | laparoscopic DP |
| 35 | F/54 | abdominal discomfort | Tail | 2 | multilocular | cystic mass | NI | NI | SPDP |
| 36 | F/63 | nausea, vomitting | Tail | 12.6 | NI | mass lesion with solid and cystic component | NI | malignant tumor of the pancreas | DP |
| 37 | F/21 | abdominal pain, fever | Tail | 2.5 | multilocular | the wall of the cyst was relatively regular, thick, and enhanced | cyst: T1 iso, T2 hyper. Rim showed hyper-intensity in DWI | SPN | laparoscopic DP |
| Our case | F/33 | NS | Tail | 3 | multilocular | 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 | SPN, NET | laparoscopic SPDP |

NS: No symptoms
NI: No information
DWI: Diffusion weighted image
DP: Distal pancreatectomy
SPDP: Spleen preserved distal pancreatectomy

Therefore, obtaining pathological evidence of ECIAS using EUS-FNA appears to be rather difficult, because the amount of solid component is too small in almost all cases to be successfully biopsied by EUS-FNA. In addition, the risk of dissemination should be considered with a cystic malignant tumor.

Some studies in the literature describe the diagnostic utility of $^{99m}$TC-Sn-colloid scintigraphy for intrapancreatic accessory spleens because $^{99m}$TC-labeled colloid taken up by the splenic tissue can help achieve a specific diagnosis in the case of ECIAS (41, 42). Although, this was not performed in the present case, since we did not list ECIAS in the initial differential diagnosis, it could be a specific examination useful for obtaining a correct diagnosis in cases preoperatively suspected to be ECIAS.

We at first recognized the mass as SPN and NET in the differential diagnosis because it had clinical and imaging characteristics similar to SPN and NET, both of which can present as a solid tumor with cystic degeneration. In addition, SPN is known to have a relatively high incidence in young women's pancreatic tails. In a retrospective review of the imaging studies of the present case, although the solid component of the mass was enhanced in the early phase of dynamic CT, the densities of the solid component and spleen were very similar; furthermore, the density was slightly higher than that of the pancreatic parenchyma. On MRI, the intensity of the solid component on T1-WI and T2-WI was similar to that of the spleen and completely dif-

ferent from that of the pancreatic parenchyma. Therefore, ECIAS should be included as one of the potential preoperative diagnoses and we should consider additional examinations including $^{99m}$TC-Sn-colloid scintigraphy to differentiate ECIAS.

In conclusion, the preoperative diagnosis of ECIAS that mimics cystic tumors is relatively difficult, because the imaging features resemble other cystic tumors or solid tumors with cystic degeneration. The features on contrast-enhanced CT and MRI include a similar density and intensity between the solid component and spleen parenchyma, which could make it possible to make a correct preoperative diagnosis of ECIAS, especially in cases with a large solid component. The efficacy of EUS-FNA for the preoperative diagnosis of ECIAS should therefore be investigated, based on an accumulation of additional cases.

**The authors state that they have no Conflict of Interest (COI).**

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