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Case and Review

Laparoscopic Resection of an Epithelial Cyst in an Intrapancreatic Accessory Spleen

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Keywords

Epithelial cyst · Intrapancreatic accessory spleen · Laparoscopy

Abstract

An epithelial cyst in an intrapancreatic accessory spleen (ECIAS) is rare. We herein report a case of a patient with ECIAS who underwent laparoscopic surgery. A 57-year-old woman was referred to our hospital because of a pancreatic tail tumor. She was asymptomatic, and a physical examination revealed no remarkable abnormalities. The levels of the tumor marker carbohydrate antigen 19-9 (CA19-9) and s-pancreas-1 antigen (SPan-1) were elevated. Ultrasonography showed a well-defined homogeneous cystic tumor. Computed tomography showed a well-demarcated cystic tumor in the pancreatic tail. Magnetic resonance imaging showed that the cystic tumor exhibited low intensity on T1-weighted images and high intensity on T2-weighted images. The cystic tumor was diagnosed as mucinous cystic neoplasm preoperatively. The patient underwent laparoscopic spleen-preserving distal pancreatectomy. A histopathological examination revealed the cyst wall to be lined by stratified squamous epithelium within splenic parenchyma, and the ultimate diagnosis was ECIAS. The postoperative course was uneventful, and the patient was discharged on postoperative

day 12. ECIAS is very difficult to diagnose preoperatively. Laparoscopic surgery is a safe and minimally invasive procedure for patients with difficult-to-diagnose pancreatic tail tumor suspected of having low-grade malignancy.

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Background

An accessory spleen is not rare and is observed in 10% of patients at necropsy [1], and 17% of accessory spleens are located within the pancreatic tail [2]. However, an epithelial cyst in an intrapancreatic accessory spleen (ECIAS) is rare. It is very difficult to diagnose preoperatively using conventional imaging such as ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI). Therefore, ECIAS is commonly misdiagnosed as other cystic neoplasms, and patients undergo surgical resection. Laparoscopic distal pancreatectomy (Lap-DP) is a safe and minimally invasive procedure and has recently been accepted for benign or low-grade malignant tumors located in the distal pancreas.

We herein report a case of a patient with ECIAS who underwent laparoscopic spleen-preserving distal pancreatectomy (Lap-SPDP) and include bibliographical comments.

Case Presentation

A 57-year-old woman was referred to our hospital because a pancreatic tail tumor had been detected during a health examination. She was asymptomatic, and a physical examination revealed no remarkable abnormalities. A laboratory examination showed normal findings. The levels of the tumor marker carbohydrate antigen 19-9 (CA19-9) and s-pancreas-1 antigen (SPan-1) were elevated to 439 U/mL (normal range, <37 U/mL) and 160 U/mL (normal range, <30 U/mL), respectively. US showed a well-defined homogeneous cystic tumor (Fig. 1a). CT showed a well-demarcated cystic tumor measuring 2.2 cm in size in the pancreatic tail (Fig. 1b). MRI showed that the cystic tumor exhibited low intensity on T1-weighted images and high intensity on T2-weighted images (Fig. 1c, d). Based on these preoperative examination findings, the pancreatic tumor was diagnosed as mucinous cystic neoplasm.

The patient underwent Lap-SPDP. Under general anesthesia, the patient was placed in the right semilateral position. Three trocars were placed: (1) at the middle umbilical site for laparoscopy (12 mm), (2) at the epigastric margin as a working trocar (5 mm), and (3) at the left lateral abdomen on the anterior axillary line as a working trocar (12 mm) (Fig. 2a). Laparoscopic US was performed to confirm the location of the tumor and to determine the resection line of the pancreas. Dissection was performed within the avascular plane along the posterior surface of the pancreas from the inferior border. Many branches of the splenic vessels were divided from the pancreatic body toward the tail using a vessel-sealing device, and the distal pancreas was mobilized (Fig. 2b). The pancreatic parenchyma was transected with an endoscopic linear stapler. After enlarging the middle umbilical site port, the specimen was removed from the abdominal cavity via an endoscopic bag retrieval system. A

closed drain was placed at the stump of the pancreas. The operative time was 144 min, and the intraoperative blood loss was 10 mL.

The resected specimen showed a well-demarcated cystic lesion surrounded with spleen-like tissue (Fig. 3a). A histopathological examination revealed the cyst wall to be lined by stratified squamous epithelium within splenic parenchyma (Fig. 3b). Based on these pathologic findings, the ultimate diagnosis was ECIAS. The postoperative course was uneventful, and the patient was discharged on postoperative day 12. Serum CA19-9 and SPan-1 levels returned to the normal range postoperatively.

Discussion

An accessory spleen is congenitally duplicated splenic tissue that is separated from the main body of the spleen and is observed in 10% of patients at necropsy [1]. The splenic hilum is the most common site of the accessory spleen, and 17% of accessory spleens are located within the pancreatic tail [2]. ECIAS is rare and was first described by Davidson et al. [3] in 1980. Kato et al. [4] summarized their experience with 39 cases of ECIAS in 2016. These cases involved 15 males and 24 females, and the median age was 48 years. All cystic tumors were located in the pancreatic tail. The cystic tumor appeared to be unilocular in 12 cases and multilocular in 21 cases. The average cystic tumor size was 4.5 cm. Wakasugi et al. [5] reported that preoperative serum CA19-9 levels were elevated in 31% of cases.

ECIAS has no malignant potential, so surgical resection for the ECIAS may not be necessary. However, all of these patients underwent surgical resection because the possibility of malignant cystic tumors could not be ruled out. Therefore, an accurate preoperative diagnosis for cystic tumor of the pancreas is important. On US, the solid component of the ECIAS shows the same echogenicity as the spleen. On CT, the cystic wall of the ECIAS shows contrast enhancement similar to that of the spleen during multiphase scans. On MRI, the solid component of the ECIAS shows the same intensity as that of the spleen, and cystic lesions commonly show low intensity on T1-weighted images and high intensity on T2-weighted images. Few previously reported cases were correctly diagnosed using conventional imaging [6, 7]. In particular, when the amount of accessory splenic tissue is small, accurate preoperative diagnosis by image findings is very difficult. In our case, ECIAS was diagnosed as mucinous cystic neoplasm preoperatively because the amount of accessory splenic tissue was small. An endoscopic US-guided fine needle aspiration (EUS-FNA) biopsy for the differential diagnosis of ECIAS was performed for 4 reported cases [4]. However, a correct pathological diagnosis was not achieved in any of the 4 reported cases, because the amount of solid component was too small to be successfully biopsied by EUS-FNA. In our case, EUS-FNA was not performed for the diagnosis of pancreatic cystic tumor. Therefore, the preoperative diagnosis of the ECIAS is very difficult at present. However, ECIAS should be considered in the differential diagnosis of pancreatic tail cystic tumors.

When it is difficult to diagnose an intrapancreatic cystic tumor, as in our case, and a malignant tumor cannot be completely ruled out, laparoscopic surgery is the better way to avoid the drawbacks of open surgery, such as increased pain and prolonged hospitalization. Lap-DP has recently been accepted for benign or low-grade malignant tumors located in the distal pancreas. There are only 12 reported cases including our own of ECIAS treated by

laparoscopic surgery in the English literature (Table 1) [4–13]. In these cases, the median age of the patients was 50 years (range, 21–67 years) and included 3 males and 9 females. The median tumor size was 2.8 cm (range, 1.5–5 cm). The median operative time was 204 min (range, 140–278 min), and the median blood loss was 50 mL (range, 10–400 mL). The median postoperative hospital stay was 12 days (range, 3–21 days). In our case, it was thought that laparoscopic surgery was a safe and minimally invasive procedure for the patient because there was a short operative time, little blood loss, and no postoperative complication. Lap-DP and Lap-SPDP have been performed in 6 patients each. We performed Lap-SPDP because the immunological role of the spleen has been increasingly emphasized and the preservation of the spleen is preferable to avoid long-term complications associated with splenectomy [14].

Conclusions

ECIAS is rare and very difficult to accurately diagnose preoperatively. Laparoscopic surgery is a safe and minimally invasive procedure for patients with difficult-to-diagnose pancreatic tail tumor suspected of having low-grade malignancy.

Statement of Ethics

Approval from the ethics committee was not required for this case report. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Disclosure Statement

None of the authors has any financial conflicts of interest to declare.

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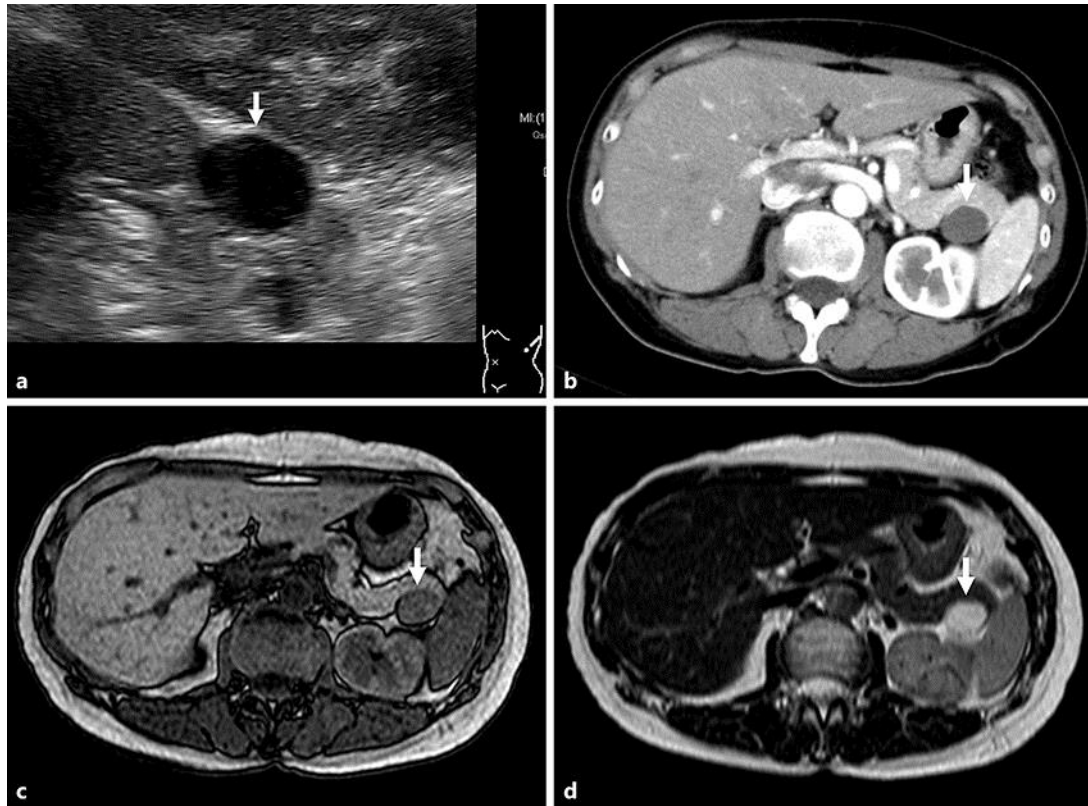


Fig. 1. **a** Ultrasonography showed a well-defined homogeneous cystic tumor (arrow). **b** Contrast-enhanced computed tomography showed a well-demarcated cystic tumor measuring 2.2 cm in size in the pancreatic tail (arrow). **c, d** Magnetic resonance imaging showed that the cystic tumor exhibited low intensity on T1-weighted images (**c**; arrow) and high intensity on T2-weighted images (**d**; arrow).

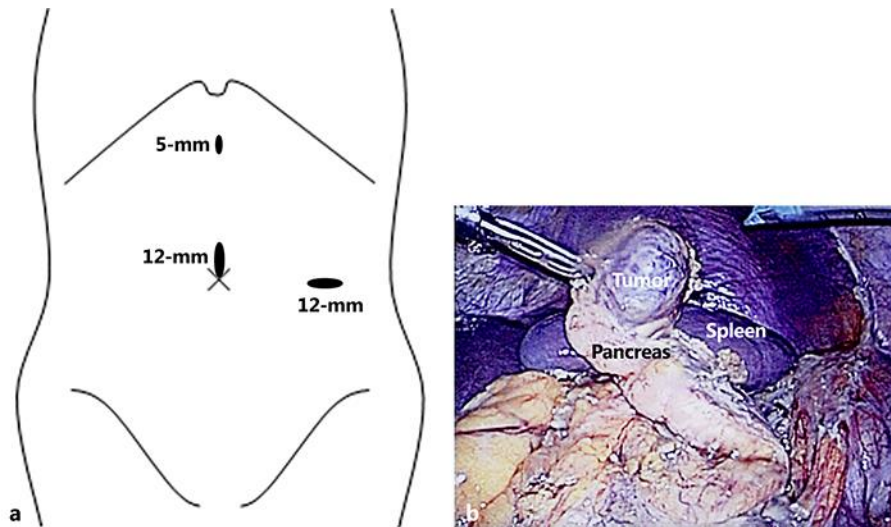


Fig. 2. a Port placement. b Intraoperative findings.

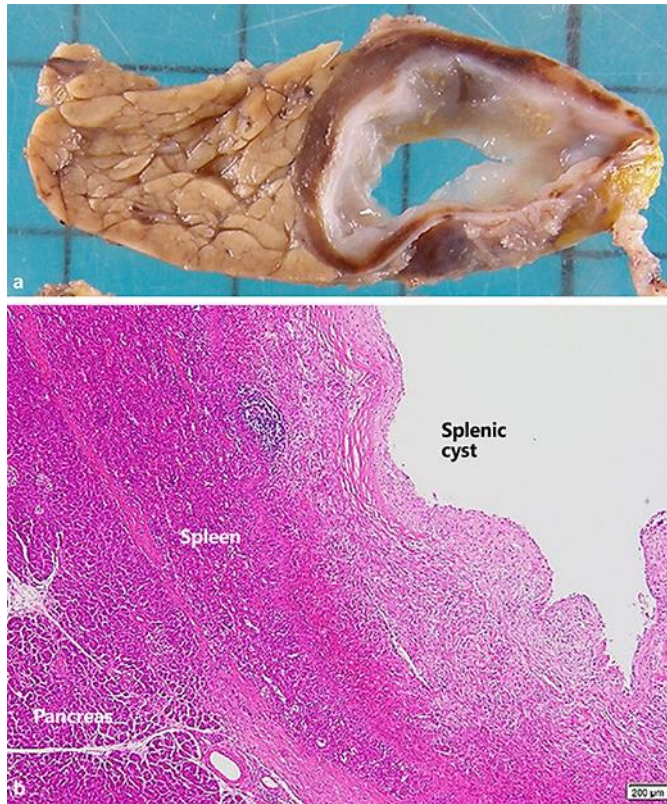


Fig. 3. **a** The resected specimen showed a well-demarcated cystic lesion surrounded by spleen-like tissue. **b** A histopathological examination revealed the cyst wall to be lined by stratified squamous epithelium within splenic parenchyma. Hematoxylin-eosin staining, $\times 40$.

Table 1. Reported laparoscopic surgical cases of an epithelial cyst in an intrapancreatic accessory spleen

No.	First author [Ref.], year	Age, years	Sex	Symptom	Location	Size, cm	Form of cyst	Preoperative serum CA19-9	Postoperative serum CA19-9	Preoperative diagnosis	Surgical procedure	Operative time, min	Intra-operative blood loss, mL	Postoperative complications	Postoperative hospital stay, days
1	Itano [6], 2010	67	M	Epigastric pain	Tail	1.5	Uni	182 U/mL	ND	ECIAS	Lap-DP	227	400	None	7
2	Iwasaki [8], 2011	36	F	Left hypochondalgia	Tail	3.4	Multi	79 U/mL	NR	MCN	Lap-DP	180	30	None	12
3	Khashab [9], 2011	49	F	Abdominal pain	Tail	2.3	ND	ND	ND	NET	Lap-SPDP	ND	ND	ND	ND
4	Urakami [7], 2011	50	F	Asymptomatic	Tail	3	Uni	ND	ND	ECIAS	Lap-SPDP	246	Minimal	None	10
5	Panagioto-poulos [10], 2012	51	M	Asymptomatic	Tail	2	ND	ND	ND	Malignant cystic tumor	Lap-SPDP	ND	ND	None	3
6	Harris [11], 2012	39	M	Asymptomatic	Tail	2.5	ND	ND	ND	Malignant cystic tumor	Lap-DP	140	250	None	8
7	Wakasugi [5], 2013	37	F	Asymptomatic	Tail	4	Multi	251 U/mL	NR	MCN, IPMN	Lap-DP	278	50	PF (grade A)	21
8	Kwak [12], 2016	21	F	Abdominal pain, fever	Tail	2.5	Multi	NR	ND	SPN	Lap-DP	ND	ND	ND	ND
9	Kato [4], 2016	33	F	Asymptomatic	Tail	3	Multi	NR	ND	SPN, NET	Lap-SPDP	ND	ND	ND	ND
10	Fujii [13], 2016	50	F	Asymptomatic	Tail	5	Uni	43.1 U/mL	NR	MCN	Lap-SPDP	ND	ND	None	14
11	Fujii [13], 2016	60	F	Back discomfort	Tail	3.5	Multi	52.9 U/mL	NR	IPMN	Lap-DP	ND	ND	None	14
12	Our case, 2017	57	F	Asymptomatic	Tail	2.2	Uni	439 U/mL	23 U/mL	MCN	Lap-SPDP	144	10	None	12

ND, not described; NR, normal range; Uni, unilocular; Multi, multilocular; ECIAS, epithelial cyst in an intrapancreatic accessory spleen; MCN, mucinous cystic neoplasm; NET, neuroendocrine tumor; IPMN, intraductal papillary mucinous neoplasm; SPN, solid pseudopapillary neoplasm; Lap-DP, laparoscopic distal pancreatectomy; Lap-SPDP, laparoscopic spleen-preserving distal pancreatectomy; PF, pancreatic fistula.