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of LEC may be possible by comprehensively evaluating its clinical and imaging findings.

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Key words: Lymphoepithelial cyst; Preoperative diagnosis; Magnetic resonance imaging

Core tip: A lymphoepithelial cyst of the pancreas is a rare benign lesion. In this study, we reviewed four cases of pancreatic lymphoepithelial cyst (LECs) to investigate the feature of LECs. We found that LEC was associated with unique characteristics on imaging findings. A preoperative diagnosis of LEC may be possible by comprehensively evaluating its clinical and imaging findings.

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Abstract

A lymphoepithelial cyst (LEC) of the pancreas is a rare benign lesion. Because patients with LEC of the pancreas have a good prognosis, it is important that these lesions are accurately differentiated from other more aggressive pancreatic neoplasms for an appropriate treatment strategy. Previous studies have reported that a definitive diagnosis of LEC often cannot be obtained based solely on the findings of preoperative imaging (e.g., Computed tomography or Magnetic resonance imaging). In this study, we reviewed four cases of pancreatic LECs to investigate the feature of LECs. We reviewed these cases with regard to symptoms, imaging findings, surgical procedures, and other clinical factors. We found that LEC was associated with unique characteristics on imaging findings. A preoperative diagnosis

INTRODUCTION

The differentiation and classification of cystic lesions of the pancreas are important for an appropriate treatment strategy. A lymphoepithelial cyst (LEC) of the pancreas is a rare benign cystic lesion^[1,2]. It has been thought to be difficult to differentiate LEC from other pancreatic lesions such as serous cystic neoplasms (SCNs), mucinous cystic neoplasms (MCNs) and intraductal papillary mucinous neoplasms (IPMNs) because the appearance on imaging of LEC varies from patient to patient and sometimes similar to other pancreatic lesions^[3]. In many patients, the lesion is surgically resected because a neoplastic cyst cannot

Table 1 Patient demographics, clinical findings

	Year	Age	Gender	Size (mm)	Symptom	Location	CA19-9 (U/mL)
Case 1	2004	59	Male	90	Nonspecific	Body	4
Case 2	2009	49	Female	60	Nonspecific	Tail	298
Case 3	2012	56	Male	40	Nonspecific	Body	75
Case 4	2012	56	Male	60	Nonspecific	Head	96

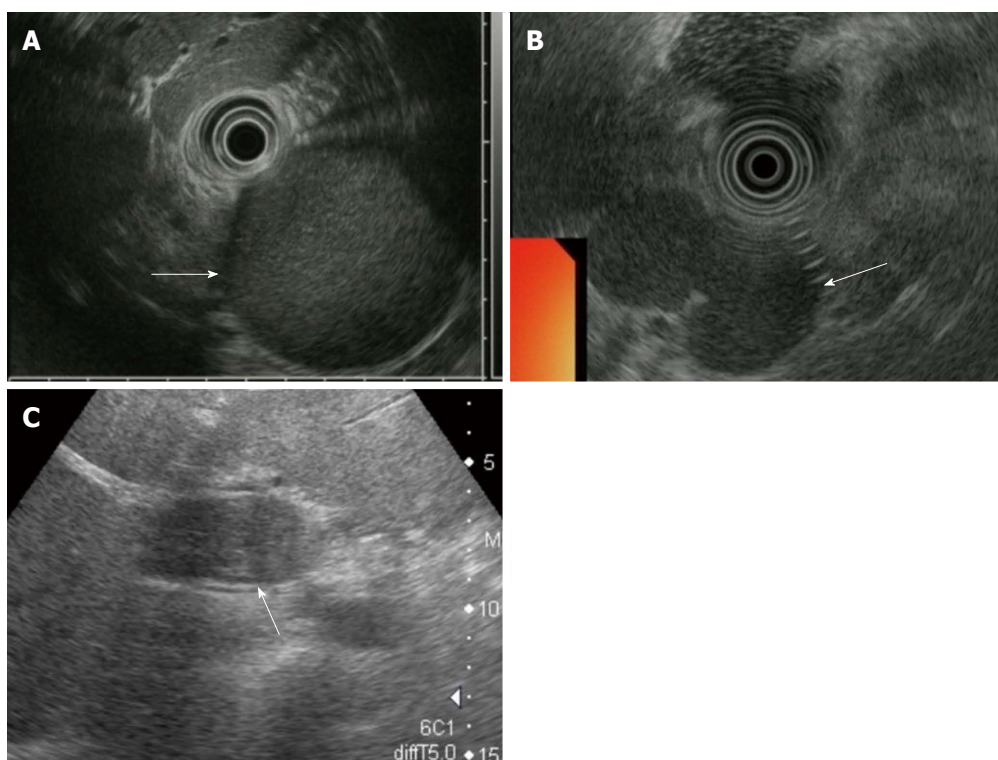


Figure 1 Ultrasonography and endoscopic ultrasonography findings. A: Findings of Patient #2, the cystic lesion had a hyperechoic appearance (arrow); B: Findings of Patient #3, the cystic lesion had a homogenous hypoechoic appearance (arrow); C: Findings of Patient #4, the cystic lesion displayed a mosaic pattern (arrow).

be ruled out^[4-6]. If a LEC can be diagnosed preoperatively, many unnecessary surgeries may be avoided. In this paper, we present four patients with LEC who underwent surgical resection and were confirmed as LEC. We summarized clinical and radiological features of LECs.

CASE REPORT

A retrospective review of our institutional database revealed four cases of LEC of the pancreas in recent 10 years. We reviewed these cases with regard to symptoms, imaging findings, surgical procedures, and other clinical factors. Various imaging studies were performed during the preoperative evaluation of the lesions. Imaging studies included abdominal ultrasonography (US), computed tomography (CT), magnetic resonance imaging (MRI), endoscopic ultrasonography (EUS) and endoscopic retrograde cholangiopancreatography (ERCP). All of the lesions were surgically resected and pathologically diagnosed as LEC.

Clinical findings

Table 1 summarized the patient demographics. Three

patients were men and one patient was a woman. The average age was 55 years. All of the lesions were detected incidentally during a work-up for unrelated reasons. In Patient #1 and Patient #3, the cystic lesions were localized in the body of the pancreas; in Patient #2, in the tail of the pancreas; and in Patient #4, in the head of the pancreas. The mean cystic size was 62.5 mm (range, 40-90 mm). Three patients had a multilocular cystic appearance and one patient had a unilocular cystic appearance. Three patients had elevated serum CA19-9 levels.

US and EUS findings

The cystic lesion in Patient #2 had a slightly hyperechoic appearance. The cystic lesion had a homogenous hypoechoic appearance in Patient #3. The cystic lesion in Patient #4 displayed a mosaic pattern. None of the three patients had solid portions the cysts (Figure 1).

CT findings

All lesions were well-defined and were exophytic off the pancreatic parenchyma. The lesion in Patient #2 had a unilocular cystic appearance, whereas the lesions in

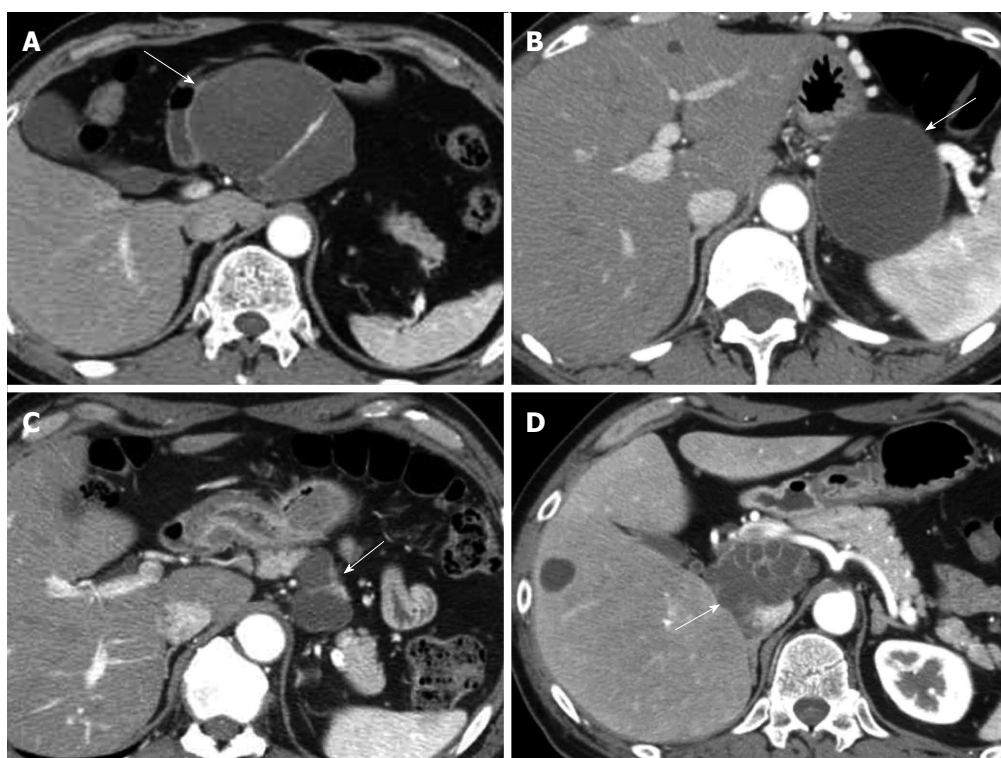


Figure 2 Computed tomography findings. All lesions were well-defined and were exophytic off the pancreatic parenchyma. The wall and septum of the cysts were enhanced. A: Findings of Patient #1, the lesion was localized in the body of the pancreas and had a multilocular cystic appearance (arrow); B: Findings of Patient #2, the lesion was localized in the tail of the pancreas and had a unilocular cystic appearance (arrow); C: Findings of Patient #3, the lesion was localized in the body of the pancreas and had a multilocular cystic appearance (arrow); D: Findings of Patient #4, the lesion was localized in the head of the pancreas and had a multilocular cystic appearance (arrow).

the other patients had a multilocular cystic appearance. The wall and septum of the cysts were enhanced. The contents of the cysts seemed homogeneous low density without enhancement. There were no solid portions within the cysts, calcification of the wall of the cyst, or dilatation of the main pancreatic duct, in any of the patients (Figure 2).

ERCP findings

All patients had normal pancreatic ducts. No patients had a communication between the pancreatic duct and the cystic lesion.

MRI findings

Figures 3-6 present the MRI findings of four patients. We presented MRI imaging of a case of SCN which had similar signal intensity with free water for comparing with the four cases (Figure 7). T1-weighted imaging of four patients showed a higher intensity than that of SCN. T2-weighted imaging and MRCP of four patients showed a lower intensity than those of SCN. Diffusion-weighted imaging (DWI) showed a higher intensity for cystic lesions than for the SCN. In particular, on DWI, the cystic lesions showed high intense signal in the central part and iso-intense in the periphery. Signal reduction in out-of phase and in-phase was not occurred in all patients.

DISCUSSION

A LEC is a rare benign lesion, which is lined with mature

keratinizing squamous epithelium and surrounded by lymphoid tissue. It typically develops in middle-aged and elderly men, and it is localized to all parts of the pancreas with equal frequency. The mean size of these cysts is 47 mm. The cyst may be multilocular (60% of patients) or unilocular (40% of patients)^[7]. Many patients with LEC have elevated serum levels of CA19-9^[8-10]. The cyst contents may vary from serous to caseous-like, depending on the degree of keratin formation^[2].

Because LEC is a benign lesion, it is often possible to select conservative treatment for ones without any significant symptoms if they can be diagnosed correctly^[11]. However, surgical resection is still commonly performed because it is difficult to distinguish them from other cystic lesions that require surgical intervention on account of their malignant potential^[4-6]. EUS-guided biopsy coupled with biochemical/tumor marker studies have recently helped to improve the diagnostic accuracy of pancreatic cysts^[12,13]. However, EUS-guided biopsy for cystic lesions of the pancreas is not generally performed in Japan because of the risk of the dissemination of tumor cells or the development of pseudomyxoma. Therefore, a preoperative pathological diagnosis is difficult and imaging studies are very important for treating cystic lesions of pancreas in Japanese.

We summarized the characteristics of LEC obtained from our cases and previous reports in Table 2.

A LEC typically develops in middle-aged and elderly men^[2]. Many patients with LEC have elevated serum CA19-9^[8-10].

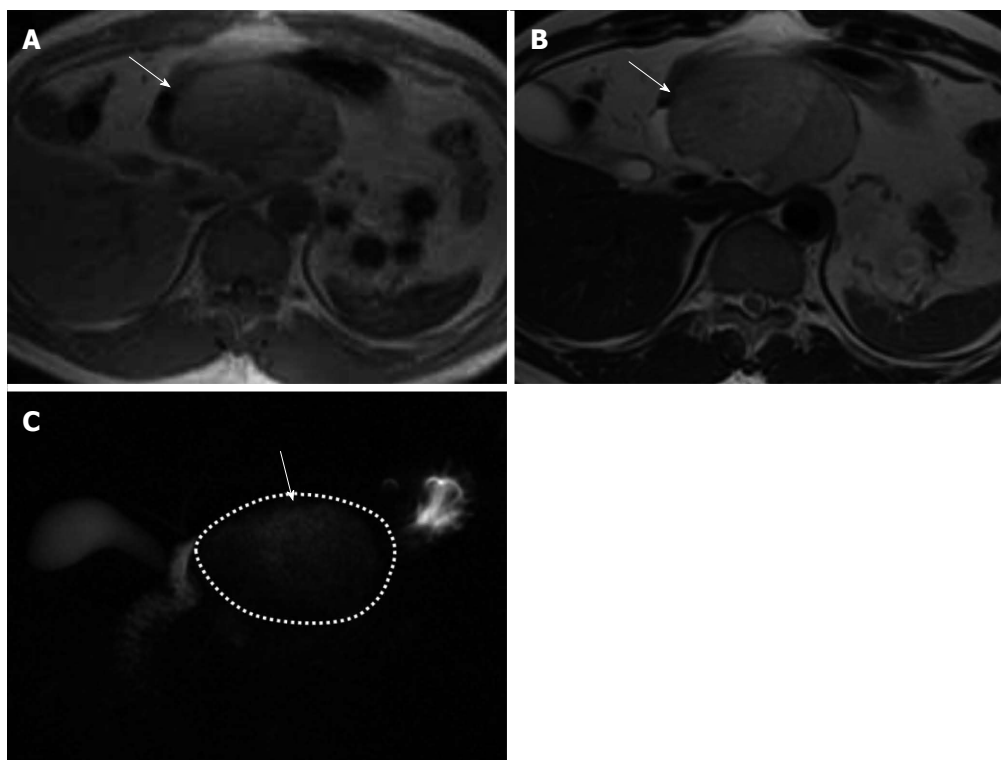


Figure 3 Findings of patient #1. A: Findings of T1-weighted imaging on magnetic resonance imaging (MRI). T1-weighted imaging of patient showed a higher intensity than free water (arrow); B: Findings of T2-weighted imaging on MRI. T2-weighted imaging of patient showed a lower intensity than free water (arrow); C: Findings of magnetic resonance cholangiopancreatography (MRCP). MRCP of patient showed a lower intensity than free water (arrow).

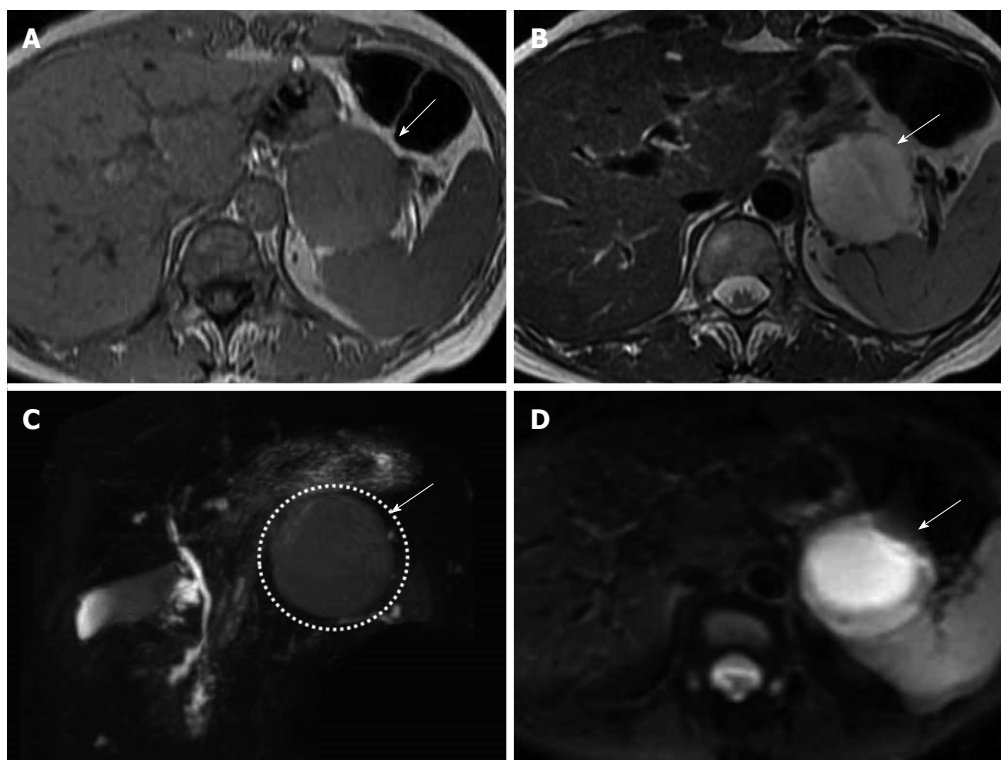


Figure 4 Findings of patient #2. A: Findings of T1-weighted imaging on magnetic resonance imaging (MRI). T1-weighted imaging of patient showed a higher intensity than free water (arrow); B: Findings of T2-weighted imaging on MRI. T2-weighted imaging of patient showed a lower intensity than free water (arrow); C: Findings of magnetic resonance cholangiopancreatography (MRCP). MRCP of patient showed a lower intensity than free water (arrow); D: Findings of diffusion-weighted imaging on MRI (arrow). Diffusion-weighted imaging of three patients showed a higher intensity than free water. The cystic lesions showed high intense signal in the central part and iso-intense in the periphery.

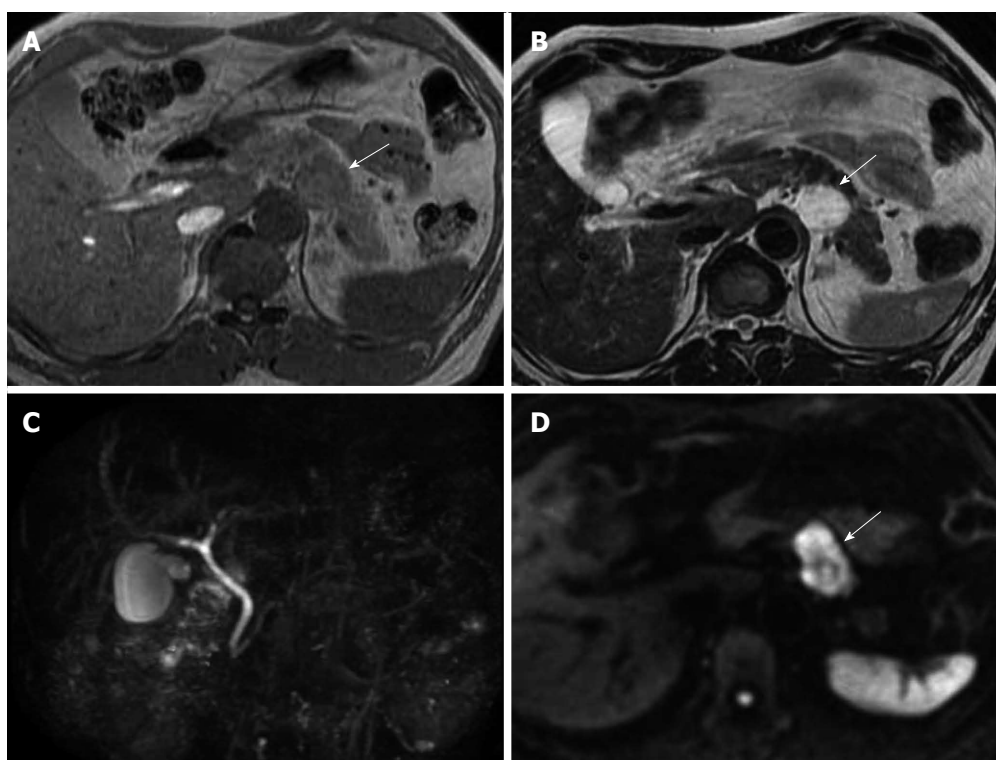


Figure 5 Findings of patient #3. A: Findings of T1-weighted imaging on magnetic resonance imaging (MRI). T1-weighted imaging of patient showed a higher intensity than free water (arrow); B: Findings of T2-weighted imaging on MRI. T2-weighted imaging of patient showed a lower intensity than free water (arrow); C: Findings of magnetic resonance cholangiopancreatography (MRCP). MRCP of patient showed a lower intensity than free water; D: Findings of diffusion-weighted imaging on MRI. Diffusion-weighted imaging of three patients showed a higher intensity than free water (arrow). The cystic lesions showed high intense signal in the central part and iso-intense in the periphery.

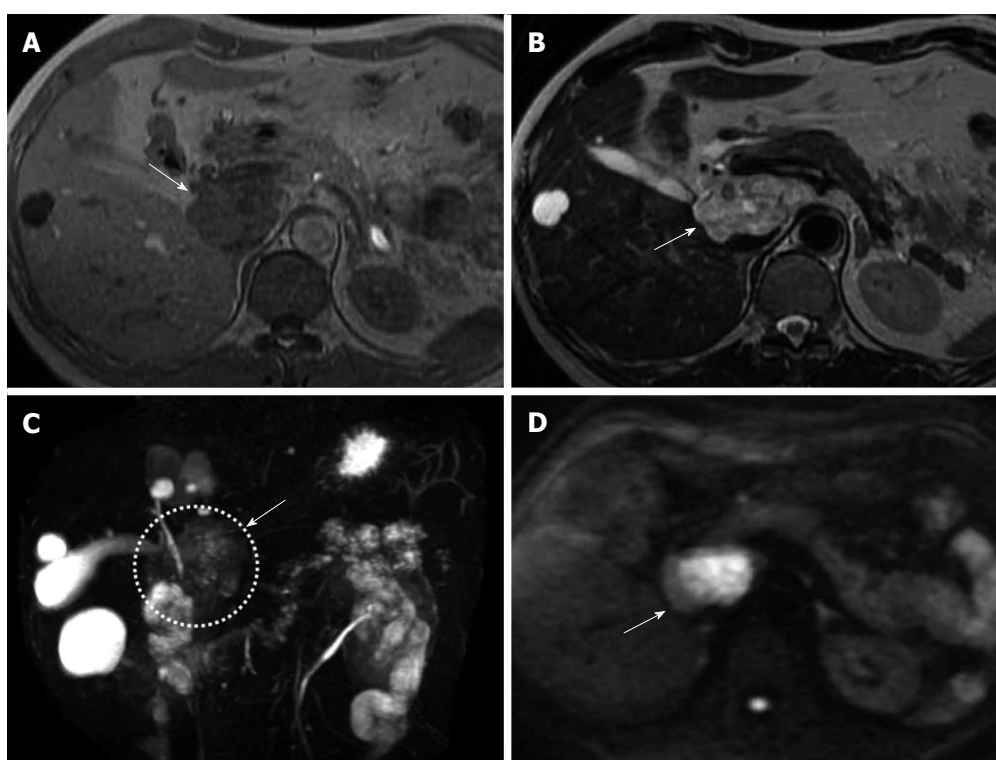


Figure 6 Findings of patient #4. A: Findings of T1-weighted imaging on magnetic resonance imaging (MRI). T1-weighted imaging of patient showed a higher intensity than free water (arrow); B: Findings of T2-weighted imaging on MRI. T2-weighted imaging of patient showed a lower intensity than free water (arrow); C: Findings of magnetic resonance cholangiopancreatography (MRCP). MRCP of patient showed a lower intensity than free water (arrow); D: Findings of diffusion-weighted imaging on MRI (arrow). Diffusion-weighted imaging of three patients showed a higher intensity than free water. The cystic lesions showed high intense signal in the central part and iso-intense in the periphery.

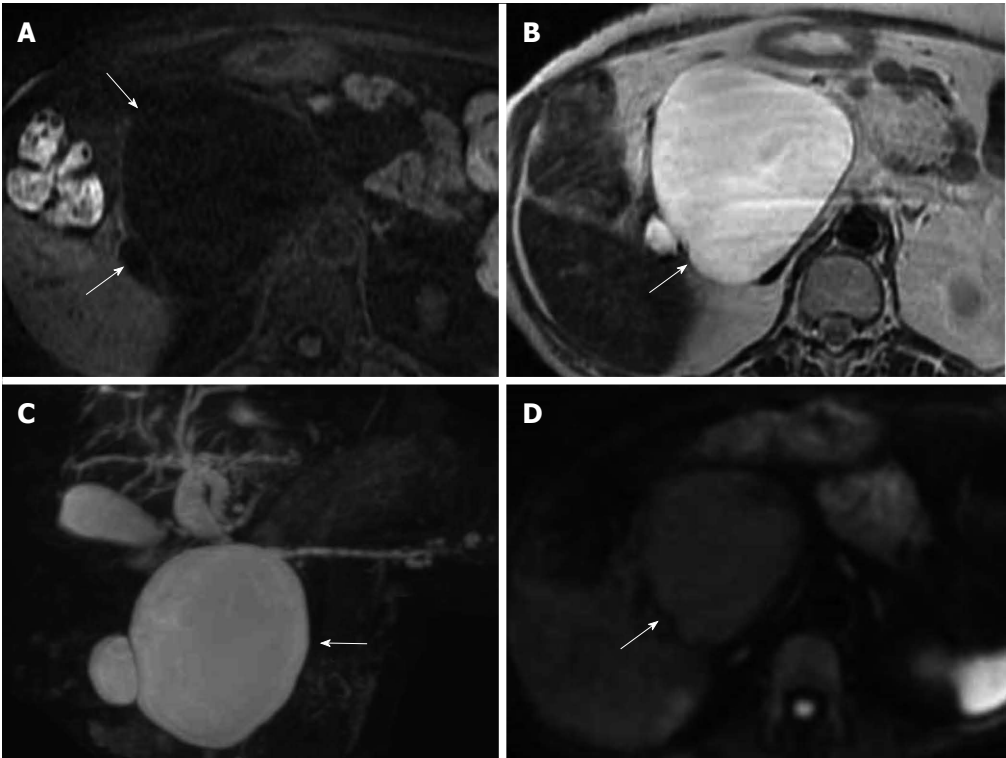


Figure 7 Findings of a case of serous cystic neoplasm. A: Findings of T1-weighted imaging on magnetic resonance imaging (MRI), the cystic lesion showed low intensity (arrow); B: Findings of T2-weighted imaging on MRI, the cystic lesion showed high intensity (arrow); C: Findings of magnetic resonance cholangiopancreatography, the cystic lesion showed high intensity (arrow); D: Findings of diffusion-weighted imaging on MRI, the cystic lesion showed high intensity (arrow).

Table 2 Feature of lymphoepithelial cyst of the pancreas

Characteristics	
Age, gender	Middle-aged and elderly men
Laboratory date	Elevation of serum CA19-9 level
The form	Well-defined Exophytic off the pancreatic parenchyma Multilocular (60%) or Unilocular (40%)
US findings	Mosaic pattern, depending on the degree of keratin formation
CT findings	Enhancement of the wall and septum of the cyst Low density cystic lesion without enhancement No pancreatic duct dilatation
MRI findings	
T1-weighted	Higher intensity than water
T2-weighted	Lower intensity than water
MRCP	Lower intensity than water No communication with the main pancreatic duct
Diffusion-weighted	Higher intensity than water

US: Ultrasonography; MRI: Magnetic resonance imaging; CT: Computed tomography; MRCP: Magnetic resonance cholangiopancreatography.

The form of a cystic lesion was well-defined and was exophytic off the pancreatic parenchyma. It might be multilocular or unilocular^[12].

The US or EUS findings sometimes displayed a mosaic pattern, depending on the degree of keratin formation^[5,13,14].

The CT findings demonstrated enhancement of the wall and septum of the cyst. The cyst itself showed uni-

form low density without enhancement^[12]. The cyst contained no solid portions within it.

The MRI findings of the four patients we reviewed were characteristic of LEC, when comparing the intensity of the lesion with that of free water. Many cystic tumors have an intensity that is similar to that of free water on MRI. In contrast, T1-weighted imaging of LEC showed a higher intensity than that of free water because the content of LEC included keratin formation. T2-weighted imaging and MRCP showed lower intensity than that of free water^[15]. Diffusion-weighted imaging showed higher intensity than that of free water^[10]. In particular, on DWI, the cystic lesions showed high intense signal in the central part and iso-intense in the periphery. It seemed that the part showing high intense signal indicated keratin formation and iso-intense in periphery indicated the wall.

However, these findings should be cautiously interpreted, because MCN and IPMN can sometimes show similar signal intensity if bleeding into the cyst has occurred^[16].

In summary, the clinical and radiological findings are sufficiently characteristic of LEC to establish a preoperative diagnosis for a majority of patients of LEC. It might be possible to select conservative treatment for asymptomatic patients with LEC.

COMMENTS

Case characteristics

Four cases were incidentally detected the pancreatic tumors and performed surgery.

Clinical diagnosis

Three patients had a multilocular cystic appearance and one patient had a unilocular cystic appearance.

Differential diagnosis

Serous cystic neoplasms, Mucinous cystic neoplasms, intraductal papillary mucinous neoplasms.

Laboratory diagnosis

Three patients had elevated serum CA19-9 levels.

Imaging diagnosis

All lesions were well-defined and were exophytic off the pancreatic parenchyma.

Pathological diagnosis

All of the lesions were pathologically diagnosed as the lymphoepithelial cyst.

Treatment

All of the lesions were surgically resected.

Related reports

A lymphoepithelial cyst of the pancreas has been thought to be difficult to differentiate from other pancreatic lesions.

Experiences and lessons

The authors found that the lymphoepithelial cyst was associated with unique characteristics on imaging findings. A preoperative diagnosis of the lymphoepithelial cyst may be possible by comprehensively evaluating its clinical and imaging findings.

Peer review

The discussion is simple. In this article, there were any findings about PET-CT and something else.

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