J Med Sci 2016;36(3):126-130
DOI: 10.4103/1011-4564.185219

CASE REPORT

Successful Laparoscopic Spleen-preserving Distal Pancreatectomy for a Huge Lymphoepithelial Cyst of the Pancreas Tail in a Young Man: A Case Report and Literature Review

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Lymphoepithelial cyst (LEC) of the pancreas is rare true pancreatic cyst. The cause is unclear. We present the case of a 19-year-old man diagnosed with LEC of pancreas tail. He had experienced intermittent upper abdominal pain and distention for 2 months prior to admission. He had no any abdominal operation history. Abdomen computed tomography (CT) scan revealed a huge homogeneous mass lesion, 15cm in the long diameter, over the pancreas tail. Then he underwent laparoscopic spleen-preserving distal pancreatectomy after a series of preoperative evaluation. LEC of pancreas tail was confirmed on the basis of clinicopathological features, such as size (15 cm), benign stratified squamous epithelium atop dense lymphoid tissue. The advantages of laparoscopic resection for LEC of the pancreatic tail were discussed.

Key words: Lymphoepithelial cyst, laparoscopic surgery, pancreatic cystic tumor

INTRODUCTION

Cystic lesions of the pancreas differ from benign to premalignant or invasive carcinoma histologically. True cysts are a very small percentage of these lesions. Lymphoepithelial cyst (LEC) of the pancreas is a rare true pancreatic cyst. The cause is unclear at present. Truong *et al*. first mentioned the name of LEC of the pancreas in 1987,[1] even though it had been described earlier. Pathologically, the lesion is composed of a stratified squamous epithelial lining atop dense lymphoid tissue. Around 100 cases have been previously described.[2,3] There are some earlier reports of laparoscopic resection of small (2.5 cm, 7.8 cm, and 4.1 cm) LEC.[3-5] Here, we describe the case of a 19-year-old male with a huge LEC of pancreas tail (15 cm in diameter), and the tumor was removed successfully by laparoscopic spleen-preserving distal pancreatectomy (DP).

CASE REPORT

A 19-year-old male presented with severe upper abdominal pain and distention after meal off and on for 2 months before admission. Local tenderness and rebounding pain in the left upper quadrant abdominal region with radiated into the back and hypoactive bowel sound were noted by physical examinations. Laboratory examinations revealed an elevated white blood cell count (11,200/μL) and level of C-reactive protein (1.35 mg/dL). Tumor markers showed elevated serum level of CA19-9 (1730/μL). Abdominal ultrasound revealed a large hypoechoic mass lesion in the pancreas tail. Computed tomography (CT) scans showed a homogeneous, well-circumscribed cystic mass lesion (15 cm × 13 cm) located at the pancreas tail [Figure 1]. The patient underwent laparoscopic resection of the pancreatic tail tumor. However, no other tumors or metastases were found during the operation.

Received: August 06, 2015; Revised: February 16, 2016; Accepted: May 18, 2016
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How to cite this article: Ho MH, Lai CL, Wu SY, Chu DM, Hsieh CB, Chan DC, *et al.* Successful laparoscopic spleen-preserving distal pancreatectomy for a huge lymphoepithelial cyst of the pancreas tail in a young man: A case report and literature review. J Med Sci 2016;36:126-9.

The operative course was uneventful, and intraoperative blood loss was approximately 50 mL. The operating time was 141 min. No perioperative blood transfusion was required. At pathologic examination, a huge pancreatic cyst was measured 15 cm in size [Figure 2], and a cyst lining composed of benign stratified squamous epithelium atop a lymphoid tissue layer [Figure 3]. The pancreas also showed focal fibrosis and inflammation. The patient was discharged home on the 6th postoperative day without any complication. At 6 months follow-up, he has developed no symptoms or signs of pancreatic endocrine or exocrine insufficiency. Serum CA19-9 returned within normal range after surgery. There was no evidence of recurrence by follow-up CT scan. He was uneventful until the recent follow-up.

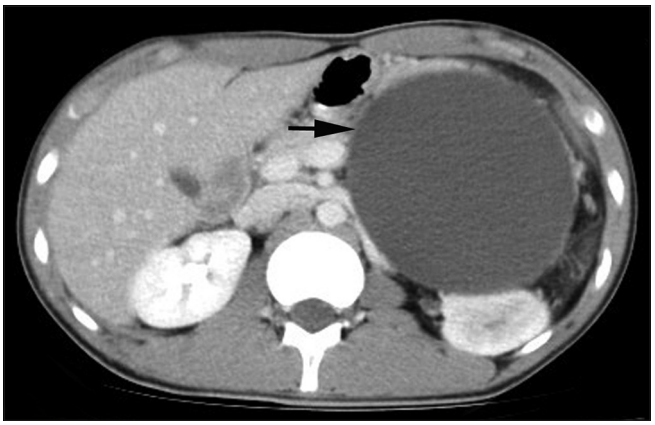

Figure 1: Computed tomography scan of the abdomen showing a large well-circumscribed homogeneous cystic mass lesion at the pancreas tail region

DISCUSSION

In this review, we searched all the cases of LEC of the pancreas published in the literature, the first case reported by Truong *et al*.[1] in 1987. Some characteristics of LEC become apparent as follow. They are seen in middle-aged patients (mean age, 55 years) predominantly in males (M/F, 4/1).[6] The most common symptom is abdominal pain, and other complaints at presentation include anorexia, weight loss, vomiting, back pain, fever, and chills. About 40% of LEC of the pancreas are asymptomatic;[5] many cases were diagnosed incidentally during the routine medical checkup or evaluation of other diseases. They can be multilocular or unilocular (in 50%),[6] and the average size is 4.1 cm. The lesion seems to be distributed over pancreatic head, body, or tail equally.[5]

Figure 2: Postoperative photograph of one large (15 cm) lymphoepithelial cyst of pancreas tail, which were resected laparoscopically, then aspirated by needle suction

Diagnosis of LEC of the pancreas preoperatively is difficult because they can mimic other cystic pancreatic lesions, and differential diagnoses of LEC of the pancreas include pseudocyst, mucinous cystic neoplasm, and intraductal papillary mucinous neoplasm.[7] Abdominal sonography can reveal LEC of the pancreas as solid masses to cystic lesions, even contrast-enhanced endoscopic ultrasound (EUS) may be useful if available.[8] The CT scan usually demonstrates features of low-attenuation mass or multilocular lesions with a thin enhancing rim. Magnetic resonance imaging on the basis of the lipid cystic component perceives a lesion with a high signal in T1 and a low signal in T2.[9] Nowadays, the EUS fine-needle aspiration cytology is the only method to make diagnosis without surgery, in spite of poor accuracy. The histological characteristics of LEC are unique, cysts lined by stratified squamous epithelium and immediately adjacent dense subepithelial lymphoid tissue that contains lymphoid follicles characterize the LEC.

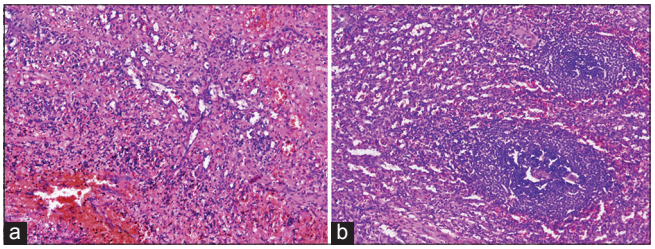

Figure 3: (a and b) Micrograph shows cyst wall lined by stratified squamous epithelium. The subepithelium shows abundant lymphocytes with germinal center formation (H and E stain, ×400)

Management of the LEC of the pancreas has been variably described in the literature, from conservative treatment with regular follow-up in asymptomatic patients to open pancreatectomy[6,10,11] or laparoscopic approach[3-5] for patients with symptoms or malignancy concern, as Table 1. No recurrences or progression into lymphoma or carcinoma has been documented in the cases of LEC in which follow-up information was available.

Because patients with LEC of the pancreas have a good prognosis, minimally invasive operation with solid

Table 1: Literature of patients with lymphoepithelial cyst underwent surgical intervention

Patient number	Author (reference)	Age/sex	Location	Size (cm)	Clinical symptoms	Surgical approach	Surgical method	Complication	Postoperative hospital stay	Follow-up
1	Au-Yong *et al*., 2004[4]	48/male	Pancreatic tail	2.5	Abdominal pain	Laparoscopic approach	Distal pancreatectomy + splenectomy	Pancreatic fistula	3	3 months later, fistula had healed
2	Toumi *et al*., 2010[3]	43/male	Pancreatic tail	7.5	Abdominal pain, night sweats, anorexia	Laparoscopic approach	Distal pancreatectomy + splenectomy	Pancreatic fistula	6	7 months later, fistula had healed No recurrence
3	Sewkani *et al*., 2010[6]	66/male	Pancreatic head	4.3	Nil	Exploratory laparotomy	Enucleation	Nil	7	N/A
4	Ibrahim *et al*., 2012[10]	5/female	Pancreatic tail	8	Abdominal pain	Exploratory laparotomy	Distal pancreatectomy + splenectomy	Nil	10	No recurrence
5	Ibrahim *et al*., 2012[10]	6/female	Pancreatic head	5	Jaundice	Exploratory laparotomy	Pancreatoduodenectomy	Blood transfusion	20	No recurrence
6	Yanagimoto *et al*., 2013[5]	53/male	Pancreatic tail	4.1	Nil	Laparoscopic approach	Distal pancreatectomy + splenectomy	Pancreatic fistula	6	N/A
7	Satoh *et al*., 2015[11]	63/male	Pancreatic body	6.5	Nil	Exploratory laparotomy	Distal pancreatectomy + splenectomy	N/A	N/A	N/A
8	Present case	19/male	Pancreatic tail	15	Abdominal pain and distention	Laparoscopic approach	Distal pancreatectomy + spleen-preserving	Nil	6	No recurrence

N/A = Not available

organ-preserving is important if pancreatic neoplasms or other malignancy could be excluded. Several reports of laparoscopic DP or robotic approach have demonstrated the safety and feasibility, particularly for premalignant tumors, with a reasonable postoperative morbidity (pancreatic fistula: 0–26%), and with more advantages than open surgery in terms of hospital stay and length of hospital stay.[5,12] There are no differences in operating time between the laparoscopic and open approaches; lower intraoperative blood loss and higher spleen preservation rate were also noted.[12]

The operative features of laparoscopic DP for LEC of the pancreas tail included in the literature, as Table 1. Inclusive of the present case, 4 cases have been reported: All in males, 3 in an adult male and 1 in a young boy. The tumors ranged from 2.5 to 15 cm in size. About 3 patients (case 1, 2 and 6) received laparoscopic DP with splenectomy. In our presented patient, our initial surgical plan is laparoscopic spleen-preserving DP if the Frozen section could exclude the possible of malignancy. Spleen-preserving was demonstrated to reduce the possibility of infectious complication and length of hospital stay for patients underwent DP.[13] The traditional spleen-preserving DP is difficult and higher operation failure rate due to severe adhesion between the tumor and splenic vessel. However, the Warshaw operation was well established to be safety and feasibility for patients underwent spleen-preserving DP[14] and lower postoperative complication rate.[15] During our operation, we smoothly separated the pancreatic tumor from the spleen and splenic vessel and removed the intact tumor meticulously. The Frozen section disclosed benign lesion by an experienced pathologist. We preserved the spleen to prevent possible postsplenectomy infection. Then, the patient returned to normal activities soon. Our patient did not have any surgical complication, and during 6 months follow-up, no symptoms or signs of pancreatic endocrine or exocrine insufficiency were noted. Serum CA19-9 returned within normal range after surgery. There was no evidence of recurrence by follow-up CT scan. He was uneventful until the recent follow-up.

CONCLUSION

The laparoscopic DP offers clear benefits in terms of reduction of operative blood loss, increase in the rate of preservation of the spleen, enhancement of recovery, and reduction in postoperative hospital stay particularly in patients with benign tumor. Here, we reported our experience of treating a huge LEC of the pancreas tail by spleen-preserving laparoscopic DP, which effectively removed the mass without any complications. We suggested that the spleen-preserving laparoscopic DP for symptomatic patients with suspicious of LEC therefore should be considered a gold standard operation.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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