Case Report

Intrapancreatic Accessory Spleen Mimicking Nonfunctional Neuroendocrine Tumor: A Case Report with Laparoscopic Resection

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Accessory spleen is a congenital anomaly often found around the splenic hilum. Intrapancreatic location has also been reported. Since intrapancreatic accessory spleen mimics pancreatic neoplasm or lymphadenopathy, it can rarely be confirmed by imaging study. We described a 71-year-old woman who visited our outpatient clinic with epigastralgia. Imaging studies later revealed a pancreatic tail space-occupying lesion which was successfully resected by laparoscopic spleen-preserving distal pancreatectomy. Pathological examination confirmed the diagnosis of intrapancreatic accessory spleen. Relevant literature on the diagnosis and treatment of accessory spleen was reviewed.

Key words: accessory spleen, laparoscopic pancreatectomy, spleen preservation

Introduction

Accessory spleen is not an unusual congenital anomaly. According to some reports, it was found in up to 10% to 30% of the general populations on postmortem examinations.[1-3] Accessory spleen arises from a failed fusion of multiple splenic primordias in the dorsal mesogastrium at the fifth week of fetal life. It is most commonly found in the splenic hilum (more than 75% of cases), followed by the pancreatic tail (about 17%).[1,4] When an accessory spleen develops within the pancreas, it might mimic a malignant lesion, neuroendocrine tumor, serous microcystic adenoma, or atypical solid and pseudopapillary neoplasm of pancreas. Literature search on the PubMed database was performed using the term "accessory spleen" between 1960 and December 2011. Clinical manifestations, diagnosis, and treatment strategies of the condition were reviewed. To date, less than 20 tissue-proven cases of intrapancreatic acces-

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sory spleen (IPAS) have been reported in the English literature. We report a patient with an IPAS mimicking a pancreatic tail tumor which was smoothly resected by laparoscopic spleen-preserving distal pancreatectomy.

Case Report

Clinical manifestation and studies

A 71-year-old woman presented to our hospital with epigastralgia for two months. With the exception of hypertension under regular medical control, she had no known history of other systemic diseases, previous abdominal trauma or operation. No body weight loss or change in appetite was noted. Physical examinations showed no remarkable finding. Panendoscopy showed mild esophagitis and gastritis, whereas abdominal ultrasonography demonstrated mild fatty liver and a cystic lesion at pancreatic tail. Laboratory data on hemogram and liver function tests were within normal limits and those on tumor markers (carcinoembryonic antigen, carbohydrate antigen 12-5 and carbohydrate antigen 19-9) were also normal. Magnetic resonance imaging (MRI) study of the abdomen later confirmed the presence of a 1.8 cm well-defined T1 hypointense and T2 hyperintense nodule at pancreatic tail (Fig. 1). Due to the possibility of malignancy without evidence of lymphatic involvement, laparoscopic surgery was performed after discussion with the patient.

Operative method

The patient was put in supine position with left side up and in a reverse Trendelenburg position. The first 10-mm camera port was inserted through a semi-circular incision above the umbilicus. After creating a pneumoperitoneum of 12 mmHg, two other 5-mm trocars were introduced at the level of the umbilicus through the left mid-axillary line and the left mid-clavicular line, respectively. Another 5-mm trocar was inserted through the linea alba at a level halfway between the xyphoid process and the umbilicus. Following the dissection of the subjacent fascia lateral to the spleen and greater omentum by using a harmonic scalpel (Autosonix, Tyco), the lesser sac was entered with visualization of the body and tail of the pancreas. After careful dissection of the inferior border of the pancreas, the body and tail were completely freed from the retroperitoneum. The splenic vessels in the posterior wall of the pancreas were identified and dissected piecemeal with coagulation of small branches of the splenic vessels. Finally, the pancreatic tail was resected by harmonic scarpel and hand-sewn with 000 polydioxanone (PDS, Ethicon). Following extraction of the specimen through the umbilical port, one silastic drain was placed in left subphrenic region. The laparoscopic operation took 4 hours and 35 minutes with 50 mL of blood loss. The patient was discharged uneventfully on post-operative day 7.

Pathological analysis

Gross pathological examination revealed a sharply delineated and encapsulated tumor measuring 1.5 × 1.2 cm, which was completely surrounded by pancreatic tissue (Fig. 2). Histological examination of the tumor showed structure identical to that of the spleen, consisting

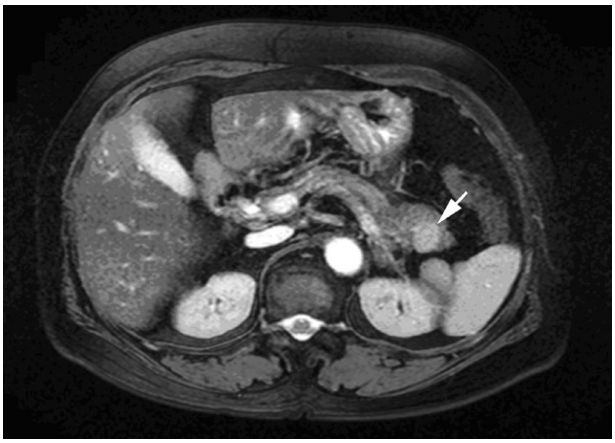

Fig. 1 T2-weighted MR imaging study showing one high-signal nodule over pancreatic tail with a size of 1.8 cm in diameter (white arrow).

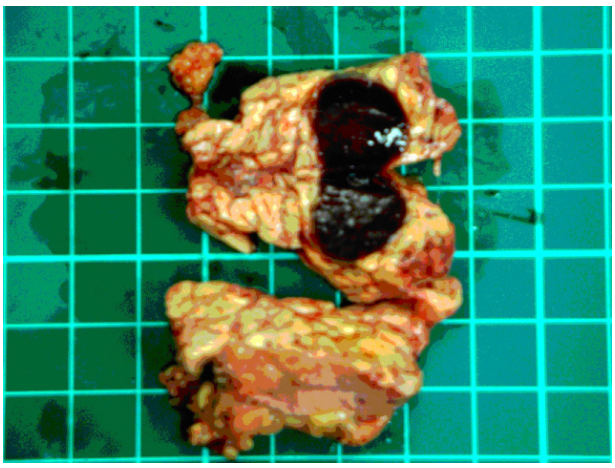

Fig. 2 Cross section of the pancreatic tail demonstrating a dark purple, sharply delineated tumor measuring 1.5 × 1.2 cm.

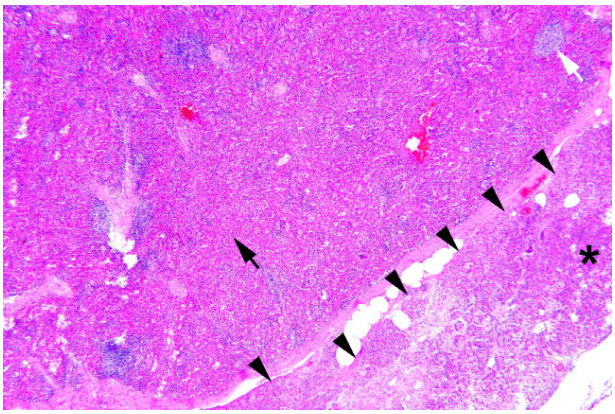

Fig. 3 Hematoxylin and eosin staining showing red (black arrow) and white (white arrow) pulps within the tumor typical of normal spleen tissue. Normal pancreatic tissue delineated with black arrowheads and exocrine acinar cells marked with asterisk.

of sinus lienalis, splenic cords, and trabecular reticulum (Fig. 3). The final diagnosis was an intrapancreatic accessory spleen.

Discussion

Ectopic spleen consists of two categories. One category is acquired splenosis arising from autotransplantation of splenic tissue, post splenectomy, or abdominal trauma-related splenic rupture, whereas the congenital category known as accessory spleen is mostly single and 1-1.5 cm in size. Although it is often found in splenic hilum, it may be located inside the pancreas (IPAS), along splenic vessels, the gastrospelnic or splenorenal ligaments, the gastric wall or intestinal wall, the greater omentum, the mesentery, the pelvic cavity and scrotum, and even in the liver. Although accessory spleen is often asymptomatic and an incidental finding that does not require special treatment, there are three circumstances under which treatment is indicated. First, when it mimics lymphadenopathy or tumor. Second, when it becomes symptomatic as in torsion, rupture and hemorrhage. Moreover, it may be removed for the treatment of hematologic disorders.[5]

Radiological differentiation between IPAS and other hypervascular tumors in the pancreas has been reported.[6] Under contrast enhanced ultrasonography, IPAS typically showed increased echogenicity compared to that of the pancreas in the arterial phase and prolonged enhancement during the portal and delayed phase.[7] In addition, most IPAS showed heterogeneous enhancement pattern at the early phase of computed tomography (CT), while it is hypointense in T1 phase and hyperintense in T2 phase in magnetic resonance (MR) images. Furthermore, Technetium-99m heat-damaged red blood cell scintigraphy typically shows hot uptake in the accessory spleen[5] and contrast-enhanced ultrasound could distinguish an IPAS from other lesions in the hepatosplenic phase.[8] In our case, MRI of the abdomen showed a 1.8 cm well-defined T1 hypointense and T2 hyperintense nodule at pancreatic tail. The picture was also compatible with that of a hypervascular metastatic tumor. However, since neither Technetium-99m heat-damaged red blood cell scintigraphy nor contrast-enhanced ultrasound was available at our institute, we could not differentiate between a malignant lesion and an accessory spleen to increase the diagnostic rate. Moreover, malignant potential of an intra-pancreatic cystic mass cannot be completely ruled out using current imaging studies.[9]

Despite current advances in radiologi-

cal technology, IPAS cannot be satisfactorily differentiated from nonfunctioning endocrine tumor or potential malignancy. Surgical intervention still remains the last resort for making the definite diagnosis. On the other hand, although management of IPAS through conventional distal pancreatectomy combined with splenectomy has been reported, splenectomy-associated complications including sepsis, venous thrombosis, cerebral infarction remain important concerns.[10] In the era of minimally invasive surgery, laparoscopic distal pancreatectomy with spleen preservation has recently been introduced. Not only does it have the advantage of spleen preservation, but it also can achieve better cosmetic results, shorter hospital stay, and earlier recovery compared with the conventional approach.[11] Laparoscopic-assisted distal pancreatectomy may be a bridge between open surgery and total laparoscopic approach. Besides, laparoscopic assistance might get better pancreatic stump control compared with the total laparoscopic approach for the beginner of laparoscopic surgery.[9] While total laparoscopic pancreatic resection with spleen preservation might be adequate for small intra-pancreatic lesions, wound extension or another incision would be necessary for removal of larger tumors. In this case, the prolonged operation was due to conversion from the initial single incision laparoscopic approach to the 4-port method due to severe adhesions between the splenic vessels and the pancreas. Although better cosmetic outcome and less wound pain may be predicted in the single-incision laparoscopic approach compared with the 4-port method, the former was impossible for our patient due to technical difficulties.

In conclusion, IPAS should be included in the list of differential diagnoses of asymptomatic pancreatic mass. Although the diagnostic rate of accessory spleen may be elevated with the aid of modern radiological tools, definite preoperative pathological diagnosis cannot be made. Laparoscopic spleen-preserving distal pancreatectomy may be a safe and feasible option in the management of pancreatic tail lesion with uncertain diagnosis.

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