Cavernous hemangioma of an intrapancreatic accessory spleen mimicking a pancreatic tumor: A case report

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Abstract

BACKGROUND
Intrapancreatic accessory spleen (IPAS) is an uncommon condition, with the majority of cases presenting as solid lesions. Thus, this condition is frequently misdiagnosed as pancreatic solid neoplasm. Moreover, splenic cavernous hemangioma is a rare disorder, whereas lesions with a cystic appearance arising from IPAS have not been reported.

CASE SUMMARY
Herein, we present a case involving a 32-year-old male who had a complex cystic lesion in the tail of the pancreas revealed by conventional ultrasound. The lesion was misdiagnosed as a pancreatic cystadenoma because of its confusing anatomic location, as well as due to its peripheral nodular and internal septal enhancement patterns on contrast-enhanced ultrasound. After multidisciplinary discussion, the patient finally underwent laparoscopic pancreatic body and tail resections. Postoperative pathology demonstrated the lesion to be a cavernous hemangioma arising from the IPAS.

CONCLUSION
Cavernous hemangioma in the intrapancreatic accessory spleen may mimic pancreatic cystadenoma, which is a condition with the potential to be malignant. Imaging follow-ups or surgical interventions may be helpful for the exclusion of malignant risks in complicated cystic lesions, especially those with parietal and septal enhancements.

Key Words: Intrapancreatic accessory spleen; Pancreas; Diagnosis; Contrast enhanced ultrasound; Case report

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Core Tip: Intrapancreatic accessory spleen (IPAS) is an uncommon condition; however, overlapping imaging manifestations of IPAS and pancreatic tumors may lead to unnecessary surgery. Cystic splenic cavernous hemangioma is a rare disorder, whereas lesions with a cystic appearance arising from IPAS have not been reported. Herein, we report a cavernous hemangioma in the IPAS that was misdiagnosed as being a pancreatic cystadenoma via contrast-enhanced modalities. The diagnosis of cystic lesions in IPAS can be challenging. Imaging follow-ups or surgical interventions may be needed for the possible malignancy risk of a complicated cystic lesion, especially those with parietal and septal enhancements.

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INTRODUCTION

An intrapancreatic accessory spleen (IPAS) is an uncommon condition, with a prevalence ranging from 1.1%-3.4% in individuals[1,2]. An IPAS is typically asymptomatic and has an innocuous nature. However, overlapping imaging manifestations of an IPAS and primary pancreatic tumors may lead to unnecessary surgery[3]. A typical IPAS demonstrates a solid lesion with a round, oval or triangular shape, which is similar to the spleen on both precontrast and contrast-enhanced images. Therefore, this disorder is frequently confused with adenocarcinomas, neuroendocrine tumors or other solid pancreatic entities. When compared with a solid IPAS, cystic lesions arising from an IPAS are rare but necessitate a differential diagnosis with pancreatic cystic neoplasms, especially those possessing the potential to be malignant. Moreover, when considering the high likelihood of false-negative results, biopsy of cystic pancreatic lesions is seldom performed, and surgery is ultimately performed in most patients.

Herein, we report such a case involving a patient who underwent laparoscopic pancreatic body and tail resections because of an indeterminate pancreatic cystic lesion. Postoperative pathology confirmed this lesion as being a cavernous hemangioma arising from an IPAS. Furthermore, the clinical and imaging characteristics of IPAS and pancreatic cystic neoplasms (according to the previous literature) were also reviewed (Table 1).

CASE PRESENTATION

Chief complaints
A 32-year-old male was referred to our hospital because of a suspicious lesion neighboring the hilum of the spleen, which was detected via conventional grayscale ultrasound in a local community hospital. The patient did not complain of obvious discomfort.

History of past illness
The patient had a history of chronic hepatitis B.

Physical examination
The patient did not complain of abdominal pain or any remarkable discomfort during the physical examination.

Laboratory examinations
In addition to a slightly increased albumin-globulin ratio (2.96) and glutamine transpeptidase level (63 IU/L), no abnormal laboratory test results, including those of related tumor markers, were found.

Imaging examinations
The patient underwent contrast-enhanced ultrasound (CEUS) in our department. Before the CEUS, a baseline ultrasound illustrated a complicated cystic nodule measuring 2 cm, with a well-defined border in the tail of the pancreas without salient blood supply on color Doppler ultrasound (Figure 1). For the CEUS, a bolus injection of the US contrast agent SonoVue (Bracco, Milan, Italy) was administered through the antecubital vein, followed by a flush of 5 mL of 0.9% normal saline. The lesion demonstrated peripheral nodular and internal septal isoenhancement in the arterial phase, followed by slight hyperenhancement of the enhanced area in the venous phase. The predominant cystic area of the lesion did not show any enhancement in either phase. According to the aforementioned enhancing pattern in the CEUS, the lesion was suspected to be a pancreatic cystadenoma via CEUS (Figure 1).
Pre-contrast and contrast enhanced ultrasound of the pancreatic lesion. A: A complicated cystic lesion (arrow) measuring 2 cm was detected in the tail of the pancreas by grayscale ultrasound in a 32-year-old male patient; B: Peripheral nodular and internal septal isoenhancement (arrow) in the arterial phase was shown on contrast-enhanced ultrasound; C and D: The enhanced part of the lesion exhibited mild hyperenhancement in the early venous phase without definite washout in the late venous phase. The cystic component did not show any enhancement through either phase.

Contrast-enhanced computed tomography (CECT) was performed to further examine the lesion. On the unenhanced CT, a nodule with a diameter of 2.2 cm and slightly low density was identified in the tail of the pancreas. Septa were observed, whereas no significant enhancement was presented within the lesion (Figure 2). The nodule was diagnosed as being a pancreatic cystic lesion via the CECT. Moreover, no salient abnormalities were found in the liver, kidney, spleen or biliary system via imaging evaluations.

**FINAL DIAGNOSIS**

The lesion was misdiagnosed as pancreatic cystadenoma by CEUS and CECT.

**TREATMENT**

After multidisciplinary discussion and communication with the patient, as well as with his family, laparoscopic pancreatic body and tail resections were performed.

**OUTCOME AND FOLLOW-UP**

Postoperative pathology demonstrated that the lesion was a splenic cavernous hemangioma in the pancreas (Figure 3). After an uneventful postoperative course, the patient was discharged on postoperative day 5. No obvious abnormality was found in a follow-up abdominal US one month later (Timeline of diagnosis and treatment of the pancreatic lesion is presented in Supplementary Figure 1).

**DISCUSSION**

Intrapancreatic accessory spleen is a rare congenital condition, compared with an accessory spleen
Table 1 Clinical and radiological characteristics of intrapancreatic accessory spleen and pancreatic cystic neoplasms

| Clinical features[1, 17,19] | IPAS | Pseudocyst | SCA | MCA | SPN | IPMN |
|-----------------------------|------|------------|-----|-----|-----|------|
| Age (mean: year)            | 40 to 65 | At any age | 60 | 40 to 50 | 30 | 65 |
| Gender                      | Slightly higher in males | Males > females | Older females | Females > males | Young females | Males > females |
| Incidence                   | 11%-17% of AS | 5%-40% after pancreatitis | 16% of PCN | 29% of PCN | 2% and 3% of PCN | 20%-50% of PCN |
| Benign/malignant            | Benign | Benign | Low malignant potential | Low malignant potential | Malignant potential |
| Anatomic location           | Tail > head/body | 1/3 near the head | Head > body/tail | Body/tail > head | Body/tail > head | Arising from the pancreatic ducts |
| Size (mean: cm)             | ≤ 2 | Depending on the duration of disease | 5-8 | 7-10 | 6 | 0.8 |
| Potential mimickers         | NET and PDAC | MCA | MCA and IPMN | MCA: IPMN and MCAC | MCA: IPMN and MCAC | SCA: MCA and MCAC |

**Radiological diagnosis**

**Ultrasound[7,17, 19-21]**

- **Baseline US**: Hypoechoic lesion with well-defined border
- **Doppler US**: Blood supply may from the splenic vessels
- **CEUS[19,21]**: Inhomogeneous hyperenhancement followed by homogeneous hyperenhancement
- **CECT[18,21-23]**: Inhomogeneous hyperenhancement followed by homogeneous hyperenhancement
- **CEMRI[22,24]**: Inhomogeneous hypointensity

**Potential mimickers**

- NET and PDAC
- MCA
- MCA and IPMN
- MCA: IPMN and MCAC
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**Radiological diagnosis**

**Ultrasound[7,17, 19-21]**

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**Potential mimickers**

- NET and PDAC
- MCA
- MCA and IPMN
- MCA: IPMN and MCAC
- SCA: MCA and MCAC
ically increased T1 signal intensity; the thickened wall shows a rim hyperintensity

| T2-W   | Homogeneous hyperintensity | The hyperintensity in tissues surrounding the pseudocyst represents the inflammation on T2 fat-suppressed images |
|--------|---------------------------|---------------------------------------------------------------------------------------------------------|
| Honeycomb pattern (microcysts) or macrocysts manifest signal intensity of simple fluid | Homogeneous high T2 signal intensity | Predominantly solid show mildly increased T2 signal intensity; cystic-dominated present T2 signal intensity closer to that of fluid |
| Papillary excrescences or nodules in the walls of the dilated ducts present hypointense on T2-weighted images |

**Management**

- Usually require no treatment
- Serial imaging follow-up
- Follow-up or resection depending on the size of the tumor
- Surgical resection

**AS:** Accessory spleen; **IPAS:** Intrapancreatic accessory spleen; **PCN:** Pancreatic cystic neoplasm; **SCA:** Serous cystadenoma; **MCA:** Mucinous cystadenoma; **SPN:** Solid pseudopapillary neoplasm; **IPMN:** Intraductal papillary mucinous neoplasm; **MCAC:** Mucinous cystadenocarcinoma; **US:** Ultrasound; **CEUS:** Contrast enhanced ultrasound; **CECT:** Contrast enhanced computerized tomography; **CEMRI:** Contrast enhanced magnetic resonance imaging; **T1-W:** T1-weighted; **T2-W:** T2-weighted.

Located at the hilum of the spleen[2,4]. Due to its innocuous nature and infrequent induction of symptoms, IPAS seldom requires therapy unless they cause symptoms as a result of the compression, torsion or spontaneous rupture of a hemorrhage[5,6].

Typical IPAS presents as a solid lesion and demonstrates similar manifestations to the spleen on both precontrast and contrast-enhanced ultrasound[7,8]. However, cystic neoplasm development in IPASs is rare. Sporadic cases of epidermoid cysts in IPASs (known as ECIPASs) have been reported[6,9-11]. The walls of ECIPASs are irregularly thickened and thicker than those of mucinous cystic neoplasms (MCNs) and intraductal papillary mucinous neoplasms (IPMNs)[9]. Moreover, the evident contrast enhancement of the partially thickened wall of ECIPAS (which is similar to that of the spleen) makes it possible to distinguish ECIPASs from MCNs or IPMNs.

The differential diagnosis was even more considerable in our case. The cystic cavernous hemangioma in the IPAS (known as CHIPAS) presented peripheral nodular and internal septal enhancements, which are frequently observed in pancreatic mucinous cystadenomas (MCAs). Furthermore, the majority of MCAs are located in the tail of the pancreas, where IPASs are also frequently discovered[12]. Therefore, this increases the difficulty of an accurate diagnosis. However, the ancillary features of a fibrous pseudocapsule or calcified contents inside of the MCNs have also been reported[13]. Another pancreatic cystic lesion that warrants vigilant discrimination from the CHIPAS is an IPMN. An IPMN in the main duct possesses a high risk of malignancy, with 38%–68% being confirmed as high-grade dysplasia or pancreatic cancer in postoperative specimens[14]. Fortunately, CEUS is sensitive in being demonstrated in the dilated main pancreatic duct and the polycystic lesion connecting to the pancreatic duct or in developing within the duct in cases of IPMNs[15].

To our knowledge, there is only one case report of solid cavernous hemangioma detected in both the spleen and the IPAS[16]. In this case, the CHIPAS was accurately identified by the investigators because of a similar enhancement pattern of the pancreatic lesion and the splenic lesions on CECT and contrast-enhanced magnetic resonance imaging. An accurate diagnosis was more difficult, as in our patient, because there was no lesion in the spleen for comparison. Moreover, a splenic hemangioma typically shows a hyperechoic and solid appearance. The atypical cystic appearance in our patient increased the
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Figure 2 Pre-operative computed tomography scan of the pancreatic lesion. A: A slightly low-density nodule measuring 2.2 cm (arrow) was found in the tail of the pancreas on unenhanced computed tomography (CT); B and C: Septa were faintly visible whereas no salient enhancement was presented within the lesion (arrows) in either the arterial or the venous phases on axial contrast-enhanced CT.

Figure 3 Hematoxylin-eosin staining of the cavernous hemangioma arising from the intrapancreatic accessory spleen. A: Large dilated vascular spaces (asterisk) separated by fibrous septa and endothelial cells (arrows) lining on the surface of the vascular spaces were observed in the intermediate-power view (original magnification, 200×); B: A high-powered photomicrograph (original magnification, 400×) illustrated splenic tissues (triangles) adjacent to the vascular spaces.

difficulty of making an accurate diagnosis.

Herein, we presented on an extremely rare case of a cystic cavernous hemangioma arising from an IPAS. Contrast-enhanced ultrasound is sensitive in demonstrating the enhancements of the septa and the parietal nodule. However, an accurate diagnosis of cystic cavernous hemangioma arising from an IPAS via imaging tools is challenging. Imaging follow-ups or surgical interventions may be needed, due to the possible malignancy risk of a complicated cystic lesion with parietal and septal enhancements.

CONCLUSION

Cavernous hemangioma in the intrapancreatic accessory spleen may mimic pancreatic cystadenoma, which is a condition with the potential for malignancy. Imaging follow-ups or surgical interventions may be helpful for the exclusion of malignant risks in complicated cystic lesions, especially those with parietal and septal enhancements.

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FOOTNOTES

Author contributions: Luo Y performed the contrast-enhanced ultrasound examination for the patient and proposed writing it up as a case report; Huang JY collected the clinical information of the patient, reviewed the literature and contributed to manuscript drafting; Yang R provided the pathological data and helped with creating the figures; Li JW contributed to revising the grammar of the manuscript; Luo Y and Lu Q were responsible for the revision of the
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