Liver Metastasis Ten Years after Excision of a Solid Pseudopapillary Neoplasm of the Pancreas

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ABSTRACT
Solid pseudopapillary neoplasm of the pancreas is a rare pancreatic neoplasm with low malignant potential. There has been only two reported cases of recurrence that occurred more than 10 years after resection of the primary tumour. We report a case of liver metastasis diagnosed 10 years after resection of a primary solid pseudopapillary neoplasm of the pancreas to illustrate the importance of long-term follow-up of this condition.

Key Words: Neoplasm metastasis; Pancreatic neoplasms

INTRODUCTION
Solid pseudopapillary neoplasm (SPN) of the pancreas is a rare pancreatic neoplasm. Despite its indolent course, malignant potential exists. We report a case of liver metastasis diagnosed 10 years after resection of primary SPN of the pancreas. To our knowledge, there have only been two reported cases of recurrence that occurred more than 10 years after resection of the primary tumour.¹,² We present this case to highlight the importance of long-term follow-up of this clinically indolent entity with the potential for recurrence and metastasis.

Case Report
A previously healthy 15-year-old boy had intermittent left-sided abdominal pain for a few years and was first admitted to the surgical unit in 2004 for increased abdominal pain lasting 1 day. Abdominal ultrasound
revealed a 9.1-cm mass with mixed echogenicity at the left upper quadrant of the abdomen. Subsequent contrast-enhanced computed tomography examination revealed a well-defined encapsulated heterogeneous mass in the left upper quadrant. Enhancing soft tissue components were present at the periphery of the lesion. Hyperdensities were present within the lesion, suggestive of recent haemorrhage. The lesion was located in the retroperitoneal cavity and impinged on the stomach, pancreas, left kidney, and spleen. The exact origin of the lesion could not be ascertained due to its large size.

Laparotomy revealed a 10-cm cystic tumour with solid component arising from the pancreatic tail. Distal pancreatectomy and splenectomy were performed. Pathological examination confirmed a SPN with widespread haemorrhagic necrosis.

After the operation, the patient was monitored clinically every 3 to 9 months. Regular follow-up ultrasound examinations at 12- to 18-month intervals were performed. No focal liver lesion was identified until 2012. Several anechoic cysts were identified in the right lobe of the liver in an ultrasound study in 2012. Follow-up ultrasound examination in 2014 revealed one of the previously detected cysts to have increased in size, from 1.1 cm to 1.7 cm. A solid component was also noted within the lesion and was suspicious of liver metastases (Figure 1a). Magnetic resonance imaging (MRI) showed a 2.4-cm heterogeneous T1 hypointense, T2 isointense-to-hyperintense lesion at segment 7 of the liver with mildly enhancing soft tissue component, suggestive of metastasis (Figure 1b). A positron emission tomography–computed tomography examination was also performed and the lesion showed mild $^{18}$fluoro-2-deoxy-D-glucose uptake (Figure 1c).

The patient subsequently underwent biopsy and percutaneous radiofrequency ablation (RFA) of the liver metastasis. Pathological examination confirmed liver metastasis from SPN of pancreas. The patient is currently well 14 months after ablation with no further evidence of recurrence (Figure 2).

**DISCUSSION**

SPN is a rare neoplasm of the pancreas, accounting for 1% to 2% of all pancreatic tumours. Overall mortality is about 2%. This rare tumour typically affects young females and has an indolent course even when distant.

![Figure 1.](image_url)
metastases are present.5

Pain and abdominal mass or fullness are the most common presenting symptoms.5 Sonographic features of the primary tumour include a well-defined hypoechoic or heterogeneous mass, sometimes with calcifications.5 Typical computed tomographic features include a well-defined cystic mass, huge mass with cystic component, or calcified cystic mass with enhancement, sometimes with calcification or haemorrhage.5 MRI usually depicts a well-defined lesion with T1 hypointensity, T2 heterogeneous hyperintensity, and an enhancing capsule.6

The recurrence rate of SPN following resection is estimated to be 10% to 15%.7 Local recurrence rate is low and is usually within 4 years of surgery.8 Metastases are reported to occur after a mean interval of 8.5 years.9 The most common site of metastasis is the liver. Regional lymph nodes, mesentery, omentum, and peritoneal metastases have also been reported.9 Kang et al10 found that tumour size larger than 8 cm, microscopic malignant features (such as cellular atypia, capsular invasion, lymphovascular invasion, perineural invasion, and peripancreatic fat invasion), and stage IV disease (peritoneal seeding and distant metastases) were significant prognostic factors for tumour recurrence. Our case had one of the above-mentioned prognostic factors — tumour size larger than 8 cm — and had subsequent development of liver metastasis after a protracted period of time.

Pancreatic SPN predominantly affects young females. A previous case series has documented a male-to-female ratio of 1:10.3 Nonetheless a more aggressive disease pattern in male patients with higher mortality and higher incidence of metastases has been suggested by a previous study11 as illustrated in our case. Therefore, long-term follow-up is necessary, especially in male patients, to detect early disease recurrence and

Figure 2. Post-radiofrequency ablation follow-up contrast computed tomography examination shows segment 7 ablation zone without evidence of tumour recurrence.
metastases.

There is currently no established guideline for management of liver metastasis. Only limited data concerning liver metastasis management are available. Most authors advocate an aggressive approach to completely remove the metastasis in view of the excellent prognosis. Surgical resection⁵ and RFA¹² of liver metastases have been reported. Case reports on treatment of liver metastasis with transarterial chemoembolisation¹³ and combined resection, followed by chemotherapy and transarterial chemoembolisation,¹⁴ are also available. Long-term follow-up is mandatory for these patients. A European expert consensus statement on cystic tumours of the pancreas recommended yearly lifelong follow-up as long as the patient remains fit for surgery.¹⁵

CONCLUSION
SPN is a rare tumour of the pancreas. Despite its indolent clinical course, malignant potential exists, even after a protracted period of time. Long-term clinical and imaging follow-up is necessary, especially in male patients. Early detection of recurrence and metastases may permit treatment by minimally invasive methods with good long-term survival.

REFERENCES