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CASE REPORT

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*

中文摘要

胰腺假乳頭狀腫瘤切除十年後出現肝轉移

林茂珠、陸國倫、黃嘉敏

胰腺假乳頭狀腫瘤是一種屬低度惡性的罕見胰腺腫瘤。文獻中只有兩宗原發腫瘤切除後十年再次復發的病例。本文報告一宗胰腺假乳頭狀腫瘤切除十年後出現肝轉移的病例，以說明對於類似病例進行長期隨訪的重要性。

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

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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.[3] Overall mortality is about 2%.[4] This rare tumour typically affects young females and has an indolent course even when distant

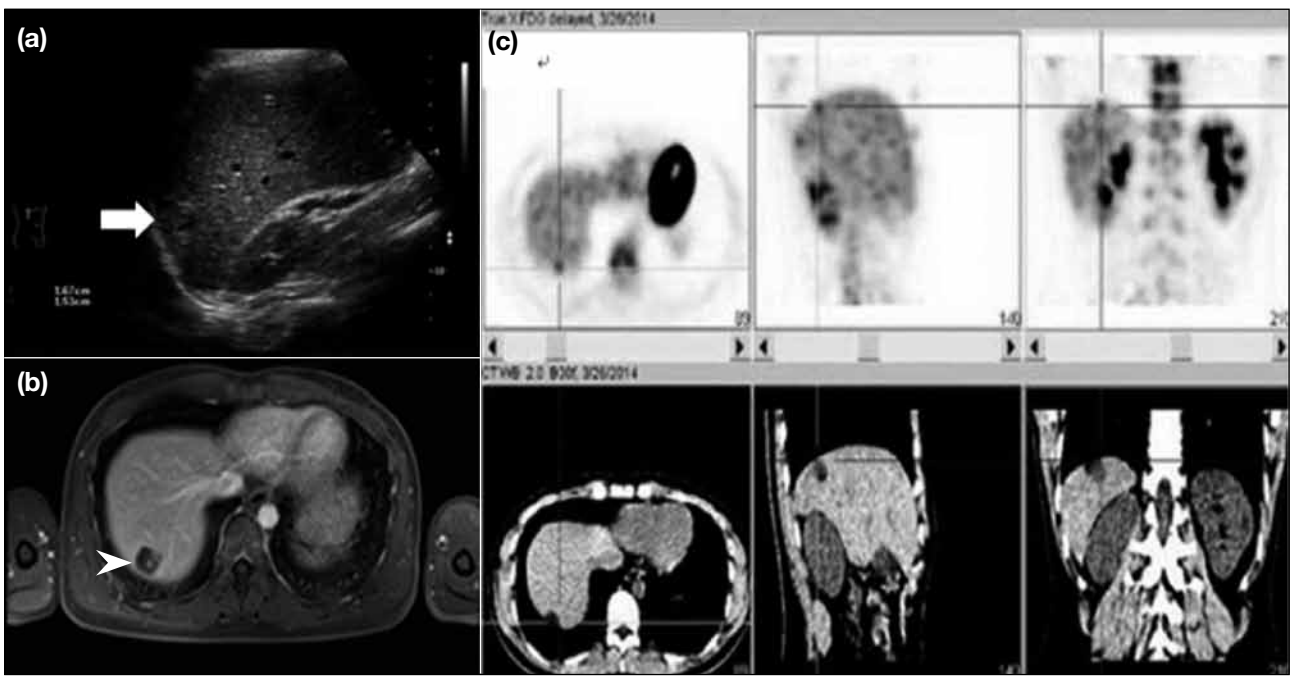

Figure 1. (a) Ultrasound: solid component is present within segment 7 cystic lesion (white arrow). (b) Magnetic resonance imaging: axial T1-weighted post-contrast image shows mildly enhancing soft tissue within the cystic lesion (white arrowhead). (c) Positron emission tomography–computed tomography: low level of ^{18}fluoro-2-deoxy-D-glucose uptake is detected within the lesion.

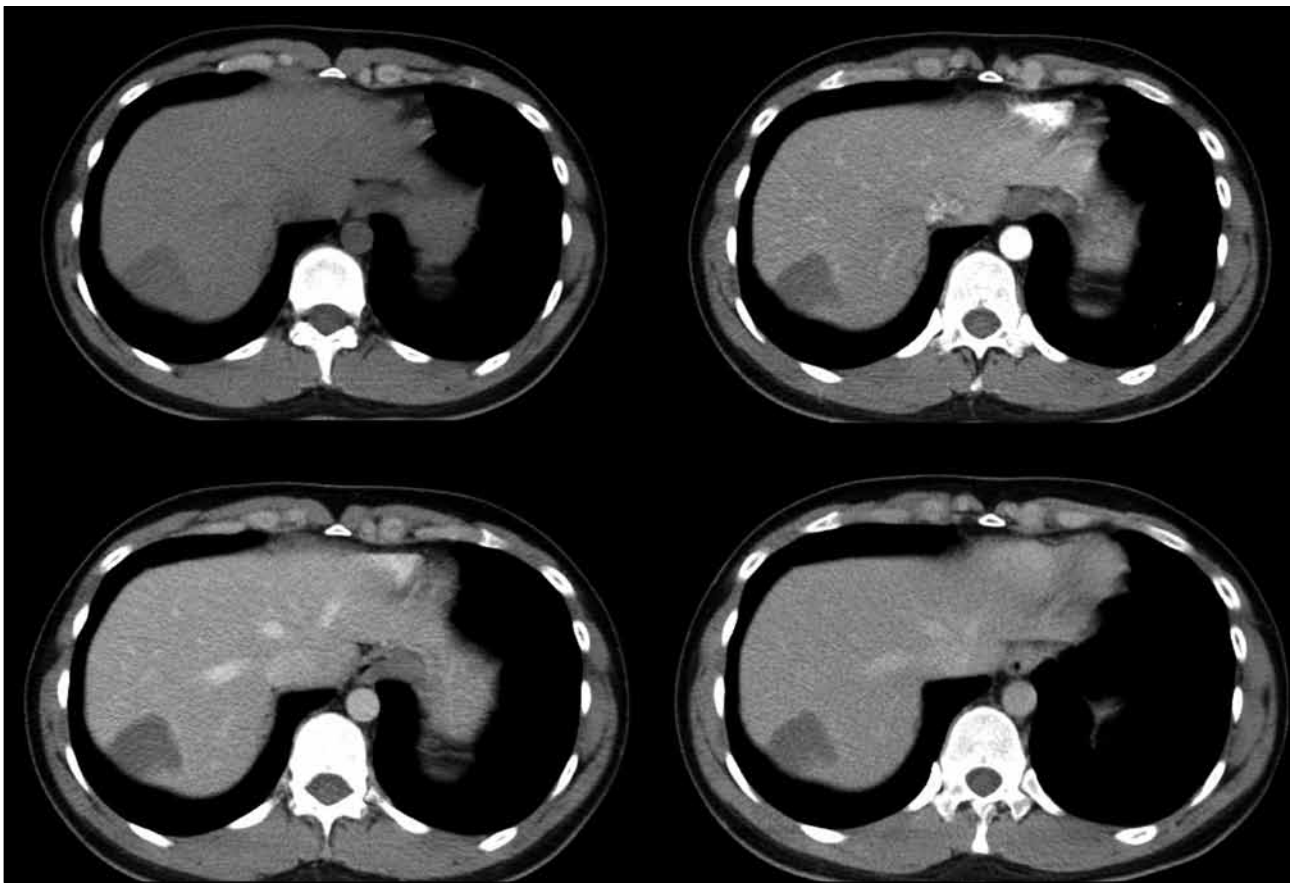

Figure 2. Post–radiofrequency ablation follow-up contrast computed tomography examination shows segment 7 ablation zone without evidence of tumour recurrence.

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 al[10] 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.[5] Nonetheless a more aggressive disease pattern in male patients with higher mortality and higher incidence of metastases has been suggested by a previous study[11] as illustrated in our case. Therefore, long-term follow-up is necessary, especially in male patients, to detect early disease recurrence and

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 resection[5] and RFA[12] of liver metastases have been reported. Case reports on treatment of liver metastasis with transarterial chemoembolisation[13] 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

1. Huang HL, Shih SC, Chang WH, Wang TE, Chen MJ, Chan YJ. Solid-pseudopapillary tumor of the pancreas: clinical experience and literature review. World J Gastroenterol. 2005;11:1403-9. cross ref
2. Gomez P, Yorke R, Ayala AG, Ro JY. Solid-pseudopapillary neoplasm of pancreas with long delayed liver metastasis. Ann Diagn Pathol. 2012;16:380-4. cross ref
3. Martin RC, Klimstra DS, Brennan MF, Conlon KC. Solid-pseudopapillary tumor of the pancreas: a surgical enigma? Am Surg Oncol. 2002;9:35-40. cross ref
4. Casanova M, Collini P, Ferrari A, Cecchetto G, Dall'Igna P, Mazzaferro V. Solid-pseudopapillary tumor of the pancreas (Frantz tumor) in children. Med Pediatr Onco. 2003;41:74-6. cross ref
5. Papavramidis T, Papavramidis S. Solid pseudopapillary tumors of the pancreas: review of 718 patients reported in English literature. J Am Coll Surg. 2005;200:965-72. cross ref
6. Guerrache Y, Soyer P, Dohan A, Faraoun SA, Laurent V, Tasu JP, et al. Solid-pseudopapillary tumor of the pancreas: MR imaging findings in 21 patients. Clin Imaging. 2014;38:475-82. cross ref
7. Geers C, Moulin P, Gigot JF, Weynand B, Deprez P, Rahier J, et al. Solid and pseudopapillary tumor of the pancreas — review and new insights into pathogenesis. Am J Surg Pathol. 2006;30:1243-9. cross ref
8. Lam KY, Lo CY, Fan ST. Pancreatic solid-cystic-papillary tumor: clinicopathologic features in eight patients from Hong Kong and review of the literature. World J Surg. 1999;23:1045-50. cross ref
9. Matsunou H, Konishi F. Papillary-cystic neoplasm of the pancreas. A clinicopathologic study concerning the tumor aging and malignancy of nine cases. Cancer. 1990;65:283-91. cross ref
10. Kang CM, Choi SH, Kim SC, Lee WJ, Choi DW, Kim SW; Korean Pancreatic Surgery Club. Predicting recurrence of pancreatic solid pseudopapillary tumors after surgical resection. A multicenter analysis in Korea. Ann Surg. 2014;260:348-55. cross ref
11. Lin MY, Stabile BE. Solid pseudopapillary neoplasm of the pancreas: A rare and atypically aggressive disease among male patients. Am Surg. 2010;76:1075-8.
12. Li JX, Wu H, Huang JW, Prasoon P, Zeng Y. Synchronous intraoperative radiofrequency ablation for multiple liver metastasis and resection of giant solid pseudopapillary tumors of the pancreas. Chin Med J (Engl). 2012;125:1661-3.
13. Prasad TV, Madhusudhan KS, Srivastava DN, Dash NR, Gupta AK. Transarterial chemoembolization for liver metastases from solid pseudopapillary epithelial neoplasm of pancreas: A case report. World J Radiol. 2015;7:61-5.
14. Tajima H, Takamura H, Kitagawa H, Nakayama A, Shoji M, Watanabe T, et al. Multiple liver metastases of pancreatic solid pseudopapillary tumor treated with resection following chemotherapy and transcatheter arterial embolization: A case report. Oncol Lett. 2015;9:1733-8. cross ref
15. Del Chiaro M, Verbeke C, Salvia R, Klöppel G, Werner J, McKay C, et al. European experts consensus statement on cystic tumours of the pancreas. Dig Liver Dis. 2013;45:703-11. cross ref