

## Whipple's procedure for an oligometastasis to the pancreas from a leiomyosarcoma of the thigh

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### Abstract

**Background** Pancreatic tumours are most frequently primary, with lesions secondary to metastasis uncommon.

**Methods** This report describes the case of a 61-year-old man who underwent resection of a right thigh leiomyosarcoma 2 years prior to presentation with obstructive jaundice. Subsequent CT and endoscopic ultrasound (EUS) diagnosed metastatic leiomyosarcoma to the pancreatic head for which he underwent a Whipple's pancreaticoduodenectomy.

**Conclusion** Metastasis from an extremity leiomyosarcoma to the pancreas is an extremely rare entity, which can be diagnosed by EUS and treated successfully by pancreaticoduodenectomy.

**Keywords** Leiomyosarcoma · Pancreas · Metastasis · Whipple's procedure

We present the case of a 61-year-old banker who presented with a 10-day history of malaise, pale, loose bowel

movements and dark urine. He had a 7 cm, high-grade, leiomyosarcoma tumour completely resected (pT2bN0R0) from his anterior right thigh 2 years previously. All surgical margins were negative for tumour. He received adjuvant external beam radiotherapy to the tumour bed (5,500 cGy over a 6-week period) and tolerated this treatment well. His past medical history was significant for ischemic heart disease for which he underwent prior coronary artery angioplasty and stenting. His laboratory investigations on presentation revealed a normal haemoglobin (13.4 g/dL) and white cell count ( $6.8 \times 10^9 \text{ L}^{-1}$ ), an albumin of 35 g/L with deranged liver function tests (total bilirubin: 133  $\mu\text{mol/L}$ , alkaline phosphatase: 831 U/L, gamma glutamyltransferase: 1,298 U/L, alanine transaminase: 649 U/L), an elevated amylase (183 U/L) and a normal CA 19-9 (47.8 kU/L). A subsequent abdominal ultrasound confirmed an obstructed biliary system with the common bile duct (CBD) dilated to 1.2 cm in diameter and a mass visible in the pancreatic head.

A CT abdomen and thorax revealed a 4.9 cm mass in the pancreatic head (Fig. 1a) which encroached the superior mesenteric artery with no evidence of pulmonary metastasis. Endoscopic ultrasound (EUS), however, revealed an encapsulated lesion in the pancreatic head, 5 cm in maximum diameter and whilst the CBD was invaded there was no involvement of adjacent vessels and no lesions suspicious for nodal or hepatic metastasis. A fine needle aspirate (FNA) was taken at that time, the cytology of which revealed spindle cells with mild cytological atypia. A subsequent endoscopic, retrograde pancreatogram demonstrated an enlarged, distorted ampulla of Vater with a malignant CBD stricture which was dilated and a plastic stent was inserted to relieve the biliary obstruction. The patient went on to have a Whipple's procedure (pylorus preserving pancreaticoduodenectomy and cholecystectomy). The histological analysis

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**Fig. 1** **a** CT image representing a  $4.9 \times 4.6$  cm area of mixed attenuation in the head of the pancreas (*arrow*). **b** High power view of metastatic leiomyosarcoma showing hypercellularity, spindle-shaped cells, moderate nuclear pleomorphism and mitotic figures (*arrows*)

of which demonstrated a 6 cm, grade III metastatic leiomyosarcoma which focally invaded the pancreas and duodenum, stained negative for c-kit and, on comparison, was identical to that resected 2 years previously from the thigh with a predominance of spindle-shaped cells (Fig. 1b). Metastatic leiomyosarcoma was diagnosed histologically. The post-operative course was uneventful and adjuvant chemotherapy and radiotherapy were deemed not to be of benefit and at latest follow-up the patient is doing well, with no complications.

The vast majority of pancreatic carcinomas are primary, and, among these, more than 90% are of ductal origin. Secondary tumours constitute 4% of pancreatic specimens [1] and often display different clinicopathological characteristics and outcomes with survival ranging from 12 to 39.6 months [2]. Metastatic pancreatic tumours are predominantly of epithelial origin, most commonly from the lung, gastrointestinal tract, kidney, breast and liver [1, 2]. Leiomyosarcoma is a high-grade malignancy originating from smooth muscle. Distant skin and soft tissue

metastases from sarcomas are very rare and often occur as a terminal event. In a series of 973 pancreatic resection specimens, only one metastatic leiomyosarcoma was found, and its origin was the retroperitoneum [1]. To date, two cases of extremity leiomyosarcoma metastasising to the pancreas have been described [3, 4]. One was a synchronous lesion identified at the time of initial presentation with a right thigh leiomyosarcoma [4] and the other presented 2 years following initial resection of a left lower leg leiomyosarcoma but had associated lung metastasis [3]. Our case represents an isolated, focal, oligometastasis of leiomyosarcoma from an extremity to the pancreas 2 years following initial resection.

The differential diagnosis between an oligometastasis and primary pancreatic carcinoma may be very difficult as signs and symptoms are similar for both primary and secondary tumours and radiological imaging is unable to differentiate the two [2]. Therefore, pre-operative diagnosis is challenging, and considerable caution is needed for patients with a previous history of non-pancreatic neoplastic disease. The spindle-shaped cytomorphologic features of leiomyosarcoma on FNA have been well-described [5]. In our case, the discovery of spindle-shaped cells on EUS-guided FNA, coupled with the background history of primary leiomyosarcoma, helped to diagnose metastatic leiomyosarcoma pre-operatively.

Metastatic tumours of the pancreas are usually diagnosed at an advanced stage, and solitary resectable metastases are infrequent. Therefore, metastatic tumours to the pancreas are rarely treated surgically. Sperti et al. [6] reported that only 3% pancreatic resections were performed for pancreatic metastases. Thus, our patient represents an extremely unusual case. As such, experience is limited and the effectiveness of surgery for these types of lesions has not been clearly established. However, metastectomy with curative intent has become standard practice for some malignancies and resection of metastatic leiomyosarcoma to other organs does confer a survival advantage if the primary site and other metastases are well controlled [7].

To conclude, this case highlights the complexity in the diagnosis of metastatic leiomyosarcoma to the pancreatic head, the role of EUS in this process and the Whipple's procedure as a treatment modality.

**Conflict of interest statement** None.

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