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A case report of primary pancreatic leiomyosarcoma requiring six additional resections for recurrences

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ABSTRACT

INTRODUCTION: Primary pancreatic leiomyosarcoma is extremely rare. We report a case in which six additional resections were required to treat recurrent tumors in a 5-year period following the primary operation.

PRESENTATION OF CASE: A 69-year-old man presented with a pancreatic tumor. Abdominal computed tomography scan showed a large heterogeneous mass with a necrotic area arising from the pancreatic body. We performed distal pancreatectomy, splenectomy, and wide resection of the transverse mesocolon. Histopathological examination confirmed the diagnosis of a pancreatic leiomyosarcoma. We repeatedly performed surgery on recurrent tumors.

DISCUSSION: Primary pancreatic leiomyosarcoma is considered to be a highly aggressive malignancy. The most effective treatment is complete surgical resection with tumor-free margins. Even when tumors recur, it is possible to improve the prognosis by further resection.

CONCLUSION: Long-term survival is achievable by repeated resection of recurrent tumors.

1. Introduction

The first report of pancreatic leiomyosarcoma was described by Ross in 1951 [1], and only 52 cases have been reported in the English literature [2–4]. The tumor is considered to derive from the vasculature or ductal smooth muscle in the pancreas [5–8]. We also discuss the clinical and histological characteristics of primary pancreatic leiomyosarcoma in a brief literature review.

The work in this case has been reported in line with the SCARE criteria [9].

2. Presentation of case

A 69-year-old man was admitted to our hospital for further examinations of a pancreatic tumor. He was asymptomatic and his physical examinations revealed no characteristic features. Except for markedly elevated glucose (205 mg/dL) and hemoglobin A1c (10.5%) levels, all laboratory findings were normal, including his tumor markers (carcinoembryonic antigen, carbohydrate antigen 19-9, Span-1, and duke pancreatic monoclonal antigen type 2). Enhanced abdominal computed tomography(CT) scan showed an 8.2×7.2 cm large heterogeneous mass with a central necrotic area in the body of the pancreas (Fig. 1a and b). Ultrasonography revealed a 9.7×7.2 cm large hypoechoic mass with cystic change in the pancreatic body (Fig. 1c), and (18F) fludeoxyglucose positron emission tomography(PET)-CT showed strong accumulation in the pancreatic body(maximum standardized uptake value: 6.12; Fig. 1d). This tumor developed markedly outside the pancreas; however, invasion into the surrounding blood vessels was not obvious. Therefore, we suspected a malignant tumor different from a conventional invasive ductal carcinoma. Our differential diagnosis was a non-functioning neuroendocrine neoplasm, acinar cell neoplasm, solid pseudopapillary neoplasm, or mucinous cystic neoplasm.

3. Primary operation

We confirmed the presence of a large tumor measuring 8 cm in its greatest diameter in the body and tail of the pancreas. The border of the tumor invaded the transverse mesocolon, and was slightly attached with the posterior wall of the stomach. The tumor was easily separable from the stomach. Therefore, we performed distal pancreatectomy, splenectomy, and wide resection of the transverse mesocolon (Fig. 2a). The cross-section of the resected tumor was a well-circumscribed whitish mass that had signs of internal hemorrhage and partial myxoid changes (Fig. 2b).

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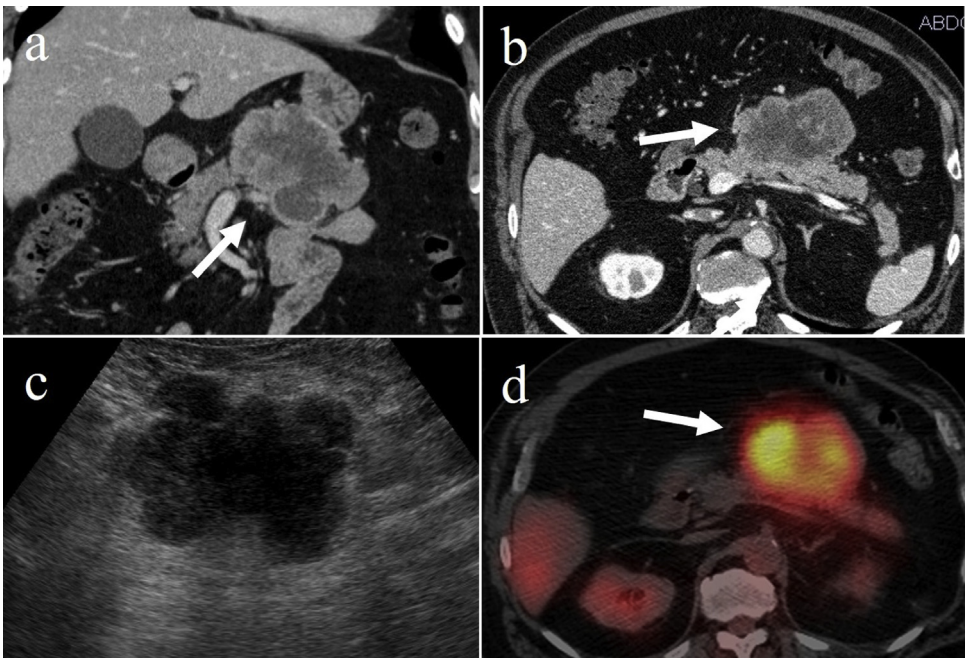

Fig. 1. Radiologic imaging modalities in the diagnosis of leiomyosarcoma.
Coronal (a) and axial (b) abdominal CT scans showing an 8.2 × 7.2 cm large heterogeneous mass with a central necrotic area in the body of the pancreas (arrows). (c) Ultrasonography showing a 9.7 × 7.2 cm large hypoechoic mass with cystic change in the body of the pancreas. (d) PET-CT showing a strong accumulation in the body of the pancreas (arrow).

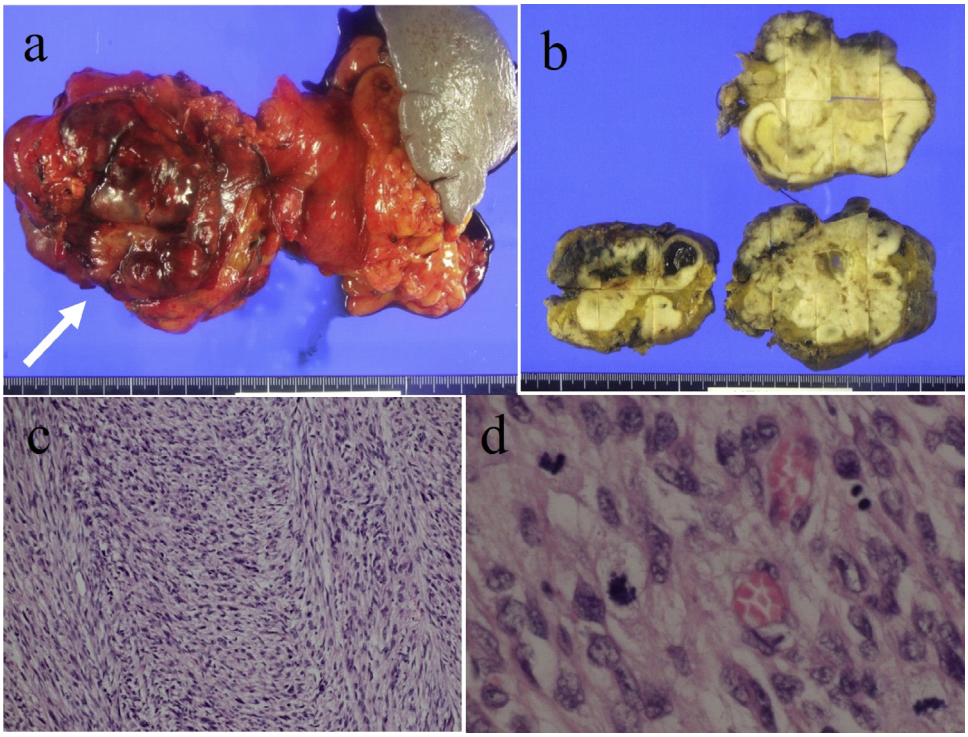

Fig. 2. Resected specimen and histological analysis.
(a) Macroscopically, there was an 8 × 7 cm tumor in the body of the pancreas (arrow). (b) The cross-section of the tumor is whitish and shows signs of internal hemorrhage and partial myxoid changes. (c) Histological examination showing interlacing bundles of spindle-shaped cells (Hematoxylin and Eosin staining x10). (d) Spindle-shaped cells with varying degrees of pleomorphism and a few mitotic figures. (Hematoxylin and Eosin staining x40).

Histological examination of the tumor revealed a composition of interlacing bundles of spindle-shaped cells with varying degrees of pleomorphic nuclei and variable mitotic activity (Fig. 2c,d). Immunohistochemical examination was positive for α-smooth muscle actin, caldesmon, and HHF35 (Fig. 3), but negative for CD117(c-kit), CD34, desmin, and S100. Given that the main muscle markers were positive, and the MIB-1 labeling index was as high as 14.5%, the tumor was diagnosed as a primary pancreatic leiomyosarcoma. Lymph node metastasis was not observed in the resected specimens. The surgical margins were negative histopath-