# **Case Report**

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# Synchronous liposarcoma of pancreas and stomach: a case report and literature review

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#### **ABSTRACT**

Liposarcoma is a common soft tissue sarcoma. However, its occurrence in pancreas or stomach is extremely rare. In the present study, a rare case of a 55-year-old female with sclerosing liposarcoma in pancreas and stomach is presented. Ultrasound, magnetic resonance imaging and computed tomography examinations were performed, which revealed a pancreatic mass. Subsequently, the patient underwent surgical resection of the tail of the pancreas and spleen and partial resection of stomach following the identification of a second mass. After surgery, the patient received no adjuvant treatment. Subsequent to 3 years of clinical follow up, the patient remains alive without recurrence or distant metastasis so far. As far as we know, this is the first reported case of sclerosing liposarcoma involving pancreas and stomach.

Keywords: Liposarcoma, Pancreas, Stomach

## INTRODUCTION

With a peak age of incidence between 50 - 65 years and a slight male predominance, liposarcoma is a common soft tissue tumor that occurs in 15 20% of sarcoma patients. However, the etiology of the tumor remains unclear. Some studies have suggested that radiation exposure or trauma may be associated with the disease development. Liposarcoma most commonly occurs in the extremities, trunk and retroperitoneum which is extremely rare in stomach or pancreas; only a small number of cases have been previously reported around the world. 1-7

In the current study, a case of synchronous sclerosing liposarcoma involving pancreas and stomach in a 55 year-old female is presented. To the best of our knowledge, this is the first case of synchronous liposarcoma in these locations.

## **CASE REPORT**

As fatigue and a poor appetite had persisted for 20 days, a 55 year old female was admitted to Shaoxing People's Hospital, Shaoxing Hospital of Zhejiang University (Shaoxing, Zhejiang, China) in May 2011. The patient had no medical history of disease and a physical examination was unremarkable. Furthermore, routine blood tests for tumor markers, carcinoembryonic antigen, cancer antigen (CA) 199, CA 125 and α fetoprotein and serum insulin levels were all within normal limits. A mass located at the body and tail of the pancreas was abdominal ultrasound. Computed identified by tomography (CT) examinations revealed that a 46x42 mm mass was located at the tail of pancreas and magnetic resonance imaging (MRI) showed a mass with stripes of hyper-density signals and well-defined margin in the T2 weighted sequence at the tail of pancreas (Figure 1).

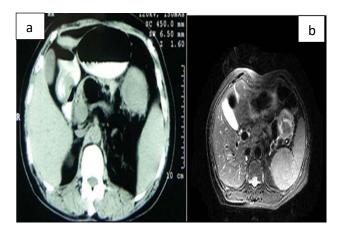


Figure 1: a) Computed tomography scan shows a lesion (46x42mm in diameter) at the tail of pancreas. b) Magnetic resonance imaging shows a mass with stripes of hyper-density signals and well-defined margin in the weighted T2 sequence at the tail of pancreas.

During a laparotomy, the indicated mass with well-defined margins was identified at the tail of pancreas (Figure 2). However, an additional mass with a diameter of 1.5cm was found at the fundus of stomach (Figure 2). Subsequently, the tail of pancreas and spleen was resected and stomach was partially resected.

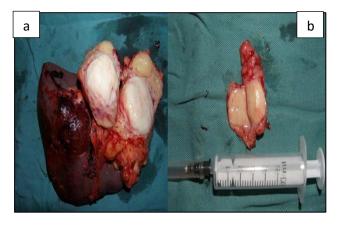


Figure 2: Macroscopic features of resected specimens. (A) A firm, well-circumscribed, white mass (4.5cm in diameter) located at the tail of pancreas. (B) A firm, well-circumscribed, white/ yellowish mass (1.5cm in diameter) located at the fundus of stomach.

Histopathological examination revealed sclerosing liposar¬coma of pancreas and stomach. The histological section showed dense collagen fibrosis associated with spindle cells (Figure 3). In addition, immunostaining revealed a marker profile (Vimentin+, S-100+, CD117-) (Figure 4).

Sclerosing liposarcoma was considered to be a low-grade malignancy and complete surgical resection was achieved with adjacent organ resection. Therefore, the patient did not undergo any adjuvant treatment and 4 years after the operation, a follow-up examination revealed that the patient was alive without recurrence or distant metastasis.

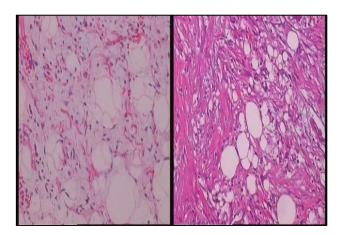


Figure 3: Histopathological features. a) Mature adipose cells mixed with spindle cells (Hematoxylin and Eosin  $\times 100$ ). b) Dense collagen fibrosis associated with spindle cells and mature adipose cells (Hematoxylin and Eosin  $\times 100$ ).



Figure 4: Vimentin and S-100 expression in the tumor as determined by immunohistochemistry. a) Vimentin expression is positive ( $\times$ 100). b) S-100 expression is positive ( $\times$ 100).

## **DISCUSSION**

As a malignant tumor of mesenchymal origin, liposarcomas are histologically subdivided into five types, including atypical lipomatous tumor (ALT)/well differentiated (WD) liposarcoma, dedifferentiated (DD) liposarcoma, myxoid liposarcoma, pleomorphic liposarcoma and mixed type liposarcoma. In addition, WD liposarcomas account for 40, 45% of all liposarcomas, which are classified into four subtypes: adipocytic (lipoma like), sclerosing, inflammatory and spindle cell WD liposarcoma. Among which adipocytic and sclerosing subtypes are the most common types of WD liposarcomas, whereas spindle cell and inflammatory subtypes are less common.8 Gastric liposarcomas are extremely rare. In 1941, the first case of liposarcoma of

stomach was reported by Abrama and Tuberville and only 11 cases have been reported in English literature so far.<sup>2,5</sup> Furthermore, only 1 case of sclerosing liposarcoma has been reported previously.<sup>5</sup> Besides, pancreatic liposarcomas are also extremely rare and the majority of cases are pancreatic metastasis.<sup>9</sup>

A case of sclerosing liposarcoma was reported by Dodo et al, in which pancreas and the second descending segment of duodenum were involved.<sup>4</sup> The histology of the two masses demonstrated a WD sclerosing liposarcoma with a dedifferentiation area within the primary mass of pancreas. WD liposarcomas exhibited no potential metastatic unless they underwent dedifferentiation.4 By contrast, no dedifferentiation was identified on histological examination in the present case. Furthermore, no invasion was observed between the two organs during surgery and the pancreatic mass was larger than that in the stomach. As most pancreatic liposarcomas are pancreatic metastases, the possibility that one of the identified masses in the current study was metastasis was ruled out. Thus, it was hypothesized that tumors located in pancreas and stomach were all primary sclerosing liposarcomas.

Notably, only 1 report regarding multiple liposarcomas has been reported in English literature with liposarcomas involving liver, subcutaneous tissue and epidural space.<sup>9</sup>

Gastroscopy is widely adopted for stomach tumors diagnosis. Endoscopic ultrasonography (EUS) was used by Yamamoto et al to diagnose liposarcoma. However, it remains difficult to differentiate between benign and malignant tumors by EUS alone, particularly when the tumor is small. CT is considered as the gold-standard technique for establishing a definite diagnosis.

However, 8 cases of gastric liposarcomas were reviewed by Seki et al and no specific features on CT scans were found.<sup>5</sup> By comparison, features of different types of liposarcomas on CT scans were reported by Tepetes et al; WD liposarcomas exhibited a heterogeneous density, myxoid types showed cystic changes and round cell and pleomorphic types were characterized by a non-specific solid structure.<sup>1</sup>

Retroperitoneal liposarcomas often exhibit non-specific symptoms. Lahat et al employed CT scans to distinguish between WD and DD retroperitoneal liposarcoma. Therefore, the presence of a focal nodular/water density area was considered to be a dedifferentiation marker. The study found that focal nodular/water density was respectively observed in 44/45 (97.8%) liposarcoma cases and 16/33 WD liposarcomas (48.5%) (P <0.001). Thus, CT scans exhibit high sensitivity to DD liposarcoma diagnosis. Nevertheless, the specificity is comparatively low. In addition, Lahat et al suggested that if CT features indicate WD liposarcoma, no further diagnostic tests are required for tumor characterization. However, a biopsy is required to differentiate between

WD and DD liposarcoma when CT features indicate DD liposarcoma.

MRI may also be utilized to identify specific histological subtypes of liposarcoma. Song et al reported that a number of MRI features may be used to differentiate histological subtypes of retroperitoneal liposarcoma.

To date, positron emission tomography (PET) has only been used for diagnosis in a small number of liposarcoma cases. Volker et al reported that for the detection of lymph node involvement and bone manifestations, PET was more accurate than conventional imaging modalities and thus PET may facilitate treatment decisions. <sup>14</sup> Moreover, Suzuki et al reported that PET was more accurate for the assessment of liposarcoma histological subtype. <sup>15</sup>

In most cases, aggressive surgical excision is considered as the gold-standard for liposarcoma treatment, as patients who undergo curative resection have been found to exhibit high survival rates. 4,16,17 A case of liposarcoma of stomach was first reported by Yamamoto et al, in which endoscopic resection was performed and no signs of recurrence were identified at 1 year after the surgery.<sup>7</sup> Among patients undergoing an incomplete excision, radiation therapy may double remission duration and possibly result in a clinical cure. However, radiation therapy has been found to exhibit no advantage in patients undergoing a complete resection.<sup>4,16</sup> Although a number of retrospective studies have reported that myxoid liposarcoma patients treated with chemotherapy exhibited a significantly higher response rate when compared with other liposarcomas, particularly DD and WD liposarcoma, liposarcomas are chemo-resistant.<sup>2,3</sup> Lahat et al reported that in their hospital, DD liposarcoma patients received systemic chemotherapy treatment, followed by an aggressive surgical approach, whereas WD liposarcoma patients were treated with less aggressive surgery alone.10 Using a less aggressive surgical approach was found to have no effect on the long term outcome of patients with WD liposarcoma. For advanced or unresectable cases, or as a supplemental post-operative therapy, chemotherapy may promising treatment.<sup>17</sup>

The prognosis of liposarcoma patients depends on the histological subtype. <sup>17</sup> Lucas et al reported the outcomes of 58 cases of WD liposarcoma with 9.3 years follow up (range: 5 months 35 years). <sup>18</sup> The cohort exhibited a recurrence rate of 53%. Furthermore, 37 (63.8%) patients were alive with no evidence of disease, 7 (12.1%) remained alive with the disease and 8 (13.8%) had succumbed to the disease (all retroperitoneal or scrotal cases). A total of 6 patients (10.3%) succumbed to other causes. Therefore, WD liposarcoma is considered to be a low-grade malignancy; metastasis will not occur without dedifferentiation. Furthermore, local recurrence may occur since it is a local disease.

In conclusion, to the best of our knowledge, the present study is the first case of sclerosing liposarcoma of pancreas and stomach reported in the literature. Due to slow growth without typical symptoms, liposarcoma is often misdiagnosed, especially when the disease is multifocal. Therefore, this also reminds us that surgeons should strengthen the exploration of other organs when handling the primary lesion.

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