Introduction

Metastatic lesions in the pancreas are rare and account for about 2% of all pancreatic malignancies (1-3). The incidence of metastatic pancreatic tumors is 1.6–11% at autopsy (4,5). Though high-volume pancreatic surgery centers are now seeing an increasing number of patients affected by pancreatic metastasis amenable to resection (6), pancreas metastasis is still unusual.

Breast cancer is one of the most common malignant tumor of woman. But breast sarcoma is more rare than breast cancer. Other than mammary alveolar cells or breast ductal cells, cells in connective tissue and adipose tissue of breast are called mammary gland stroma cells, and its malignancy is included in sarcoma (7).

Liposarcoma, defined as the malignant tumor from adipose tissue, mostly occurs in the soft tissue of retroperitoneal area or thigh and is commonly observed among adults over 40-year-old.

The pancreatic metastasis from both mammary gland stroma cell sarcoma and liposarcoma is really rare. We report two cases of pancreatic metastasis from the two types of sarcoma, both are unique.

Case presentation

Case 1

A 60-year-old woman was admitted to our hospital as ‘acute pancreatitis’. It was the second time she suffered from an acute attack in three months. The serum amylase was 252 U/L, while the uric amylase was 8,800 U/L at the checking time. After effective treatment, the pancreatitis was controlled and she recovered quickly.

However, this time the computed tomography (CT) scan identified a pancreatic mass in the tail of pancreas with length of 44 mm, width of 48 mm (Figure 1). Pancreatic carcinoma, pancreatic metastases or pancreatic pseudocyst was suspected.

On the other hand, she had undergone two operations before. One was in 2006 and the other in 2012. Pathological findings showed that the former was primary mammary gland stroma cell sarcoma, another is a 31-year-old man with mucous liposarcoma. Both of them have a history of the primary sarcoma. The imaging materials are in detail. The metastatic tumors are both resectable, and the final diagnoses of the pancreatic masses are proved through pathological section.
By her medical history, we let her take a positron emission tomography-computed tomography (PET-CT), and it showed a high-metabolic mass in the tail of pancreas, which is similar to that of CT (Figure 2). However, no other suspicious niduses were found.

At last a distal pancreatectomy with splenectomy was performed. Macroscopic examination of the operative specimen showed a 6 cm × 5 cm × 3 cm, hard mass with capsule, with invasion of starting part of jejunum and transverse mesocolon (Figure 3).

Microscopic diagnosis was a fusiform cell sarcoma in pancreas with necrosis (Figures 4, 5). Immunohistochemistry of the sarcoma cells showed, SMA (50%), Desmin (−), s-100 (−), CD34 (diffuse strongly +), CD99 (partly +), HMB45 (−), Vimentin (+), CD117 (−), PCK (−), STAT6 (−), Ki-67 (LI: 20%).

The diagnosis of pancreatic metastasis from primary mammary gland stroma cell sarcoma was clarified.

The follow-up examination half a year later than the final operation showed the patient was in good physical condition, and no nidus was found.

Case 2

A 31-year-old man presented to our hospital due to a finding of ‘pancreatic lesion’.

Over the past three years, he had three operations and even an amputation surgery on his left legs. The arch criminal was liposarcoma. The liposarcoma was first found <16.3 μg/L).

Figure 1 Epigastrium contrast-enhanced CT showed a low density in the tail of pancreas without any obvious enhancement during arterial phase.

Figure 2 PET-CT showed a hypermetabolism mass in the tail of pancreas, same position as that CT showed.

Figure 3 The excised tissue of pancreas is around 6 cm × 5 cm × 3 cm. An off-white mass with necrosis was detected. The boundary is not clear.

Figure 4 Tumor tissue (HE, ×40). ‘a’ is tissue of tumor; ‘b’ is normal interstitial tissue of pancreas; ‘c’ is normal pancreatic acinar.

Figure 5 Tumor tissue (HE, ×100).
Figure 5 Tumor tissue. Cellular proliferation of atypical spindle tumor cells is accompanied by nuclear atypia and hyperchromasia (HE, ×400).

Figure 6 Horizontal plane of epigastrium contrast-enhanced CT showed low density mass in the head of pancreas and porta hepatis.

Figure 7 Frontal plane of epigastrium contrast-enhanced CT showed low density mass in the head of pancreas and porta hepatis.

Figure 8 The biggest mass is about 4 cm × 6 cm × 6 cm. The section is off-white and soft. No capsule was found.

Figure 9 Tumor tissue. The non-staining areas among anomalous cell are adipose hollow space. Cellular proliferation of atypical spindle tumor cells is accompanied by nuclear atypia and hyperchromasia (HE, ×100).

on his left leg, then in the mediastinum. This time he found tumors on the head of pancreas and porta hepatitis by CT as follow-up examination (Figures 6, 7).

The laboratory parameters including ‘blood routine’, ‘hepatic and renal function’ and ‘tumor markers’ were all within normal levels.

An operation of ‘resection of tumor in the head of pancreas and retroperitoneal area’ was done then. Multiple hard tumors were found in the head of pancreas, porta hepatitis and retroperitoneal area. The biggest one was about 4 cm × 6 cm × 6 cm (Figure 8). Intestinal canal and other tissue beside tumors were adhesive to each other.

The pathology report proved that the tumors were mucous liposarcoma, same as before (Figure 9).
Immunohistochemistry of the sarcoma cells showed positive of s-100, MDM2 and CDK4.

Half a year later, the patient returned due to discovery of mass in retroperitoneal area. Another operation was given and the pathology report showed recurrence of the liposarcoma.

However, the patient didn’t find other nidus during last 3 months after last operation and lived a normal life.

Discussion

Sarcoma is the malignant tumor comes from mesenchymal tissues. It is less common than carcinoma and mostly occurs in younger people. The macroscopic examination of sarcoma is always nodular and lobular. As its fast growth rate, sarcoma sometimes can not only develop with the infiltrative growth pattern, but also with envelope formation and putting pressure on nearby tissue. Sarcoma is always huge, soft and with gray section.

Liposarcoma is the most usual type of retroperitoneal soft tissue sarcomas (8). However, it can only be cured by complete surgical resection (9-12). Unfortunately, R0 resection rates are only approximately 50% (10,13). Recurrence is most often identified 6 to 24 months after surgery. Recurrence is usually locoregional, rather than distant (14). Surgical resection is the primary method for locoregional control of liposarcoma.

Breast sarcoma is an infrequent but aggressive entity. Due to its rarity, there have not been sufficient studies of its clinicopathological features and adequate treatment approach to achieve a consensus in terms of management of the disease, with published articles usually being small retrospective case reports and reviews.

The primary tumors of the two cases are both unusual, as well as pancreatic metastasis. So that the two cases are unique and valuable.

Moreover, the diagnosis of pancreatic metastasis is difficult. Imaging examination like CT cannot distinguish pancreatic metastasis from pancreatic primary carcinoma clearly. Sometimes, a space between tumor and normal pancreas tissues can give a favor in differential diagnosis but not always. And pancreatic metastasis don’t have typical symptoms. Abdominal pain, jaundice and weight loss are most familiar, same as other digestive system disease. In fact, most patients don’t have any obvious discomfort. One of the differences between pancreatic metastasis and pancreatic primary carcinoma is the level of tumor markers. CA-199 and CA-125 are meaningful in amount of pancreatic carcinoma patients but are not obviously higher in pancreatic metastasis especially in metastatic sarcoma. Another importance of differential diagnosis is medical history of sarcoma. Patients with metastatic sarcoma always have exact history of the primary or metastatic sarcoma. What’s new, it is reported that high-resolution genomic profiling can be used in differential diagnosis of sarcoma (15).

Nevertheless, diagnosis of pancreatic metastasis of sarcoma still rely on medical history of sarcoma and checking mass within pancreas tissue through Imaging examination.

Traditionally, chemotherapy and radiotherapy are used as supplementary treatments before or after surgery for sarcoma. In recent years, there are now several new systemic and targeted drugs for adult soft tissue sarcomas, either approved or that show promise in clinical trials, following advances in understanding the molecular biology of sarcomas, and medical oncologists face increasingly complex treatment choices (16). However, the two patients above didn’t get any other treatment except surgical treatment.

Sarcoma is aggressive and always not once resectable to make sure not to relapse. But fortunately, the life time of sarcoma patient is much longer than pancreatic primary carcinoma. The patient of case 1 with ‘Mammary gland stroma cell sarcoma’ has been fighting against her disease over 10 years. So far as I know, an attempt at complete tumor resection can offer patients the greatest hope of long-term survival.

Acknowledgements

This work was supported by Union Hospital of Tongji Medical College of Huazhong University of Science and Technology, Wuhan, Hubei province, China.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

References

1. Sperti C, Moletta L, Patane G. Metastatic tumors to the pancreas: The role of surgery. World J Gastrointest Oncol


doi: 10.21037/jxym.2017.06.02