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Isolated metastasis of uterine leiomyosarcoma to the pancreas: Report of a case and review of the literature



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ABSTRACT

INTRODUCTION: Metastatic tumors of the pancreas are uncommon and rarely detectable clinically. Metastases to the pancreas are rare. We present a patient with pancreatic metastases from a leiomyosarcoma of the uterus and review the literature about the clinical features of pancreatic metastasis and its surgical management.

PRESENTATION OF CASE: A 40-year-old woman, who underwent hysterectomy, left oophorectomy, omentectomy and lymph node dissection for leiomyosarcoma of the uterus. At the follow up, the patient complained of non-specific abdominal discomfort. Preoperative diagnosis were pancreatic pseudocyst, cystadenoma or cystadenocarcinoma. At laparotomy, a cystic mass was found in the tail of the pancreas which was invaded to the transverse colon mesentery and the spleen. Distal pancreatectomy with splenectomy and transverse colon resection was performed. Histologically, the tumor was evaluated as poorly differentiated leiomyosarcoma.

DISCUSSION: Metastatic lesions of the pancreas are uncommon and less than 2% of all pancreatic malignancies. However a few cases of leiomyosarcoma with metastases to the pancreas have been reported in the literature. Before deciding that the lesion in the pancreas was metastasis, primary leiomyosarcoma of the pancreas had to be ruled out. Histologically, leiomyosarcoma of the pancreas contains interlacing spindle cells with varying degrees of atypia and pleomorphism. The surgical approach to the pancreatic metastases must be aimed complete excision of the tumor with a wide negative margin of clear tissue and maximum preservation of pancreatic remnant if possible.

CONCLUSION: In the absence of widespread metastatic disease, aggressive surgical approach with negative margins must be aimed.

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1. Introduction

Metastatic tumors of the pancreas are uncommon and rarely detectable clinically. The reported incidence of these tumors ranges from 3% to 11% in autopsy series.¹ Most patients with pancreatic metastases have widespread disease. Isolated, potentially resectable pancreatic metastases are detected infrequently.² Although surgical resection of metastatic lesions to the liver, brain and lungs improve survival and quality of life, the management of pancreatic metastases has not been clearly defined.^{2,3} Uterine leiomyosarcoma is a rare and aggressive malignant neoplasm of the myometrial smooth muscle cells. The most common sites

of distant metastases from leiomyosarcoma are the lung, kidney and liver. Metastases to the pancreas is rare.^{4,5} The resection of pancreatic metastases was uncommon in the past because of the high mortality and morbidity rates of pancreatic surgery. However, in recent years an improvement on pancreatic surgery has been clearly shown.⁶ We present a patient with pancreatic metastases from a leiomyosarcoma of the uterus and review the literature about the clinical features of pancreatic metastasis and its surgical management.

2. Presentation of case

A 40-year-old woman, who underwent hysterectomy, left oophorectomy, omentectomy and lymph node dissection for leiomyosarcoma of the uterus in December, 2008 and treated later with radiation therapy and chemotherapy. The patient received adjuvant chemotherapy with 75 mg/m² doxorubicin a day and

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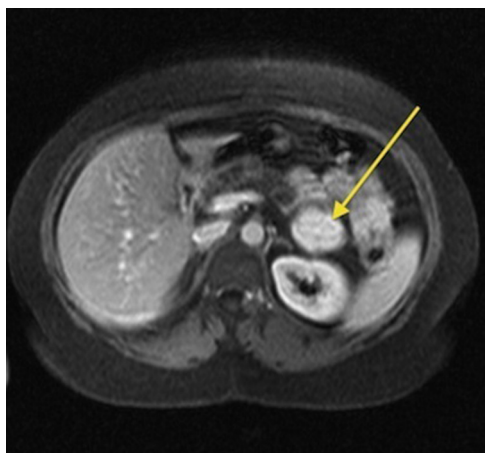


Fig. 1. Post-contrast T1-weighted MRI image of distal pancreatic mass shown with yellow arrow.

100 mg/m² cisplatin a day and received external-beam radiotherapy to the whole pelvis about 28 days. She was well without evidence of disease recurrence at follow-up to May, 2013. At this time, the patient complained of non-specific abdominal discomfort and physical examination revealed a small epigastric mass. Laboratory values revealed a border-line anemia with a hemoglobin value of 11.1 gr/dl (12.2–16.2 gr/dl), hypoalbuminemia with a albumin value of 3.1 gr/dl (3.5–5.2 gr/dl) and an increase of CA-125 with a value of 445 U/ml (2.6–24 U/ml). Abdominal ultrasonography showed a hypoecoid mass measuring 4 cm at the widest diameter in the tail of the pancreas. CT scan and MRI showed only the pancreatic mass with a 4 cm at the widest diameter in the tail of the pancreas, with no evidence of additional lesions (Fig. 1). A diagnosis of the serous papillary cystadenoma of the

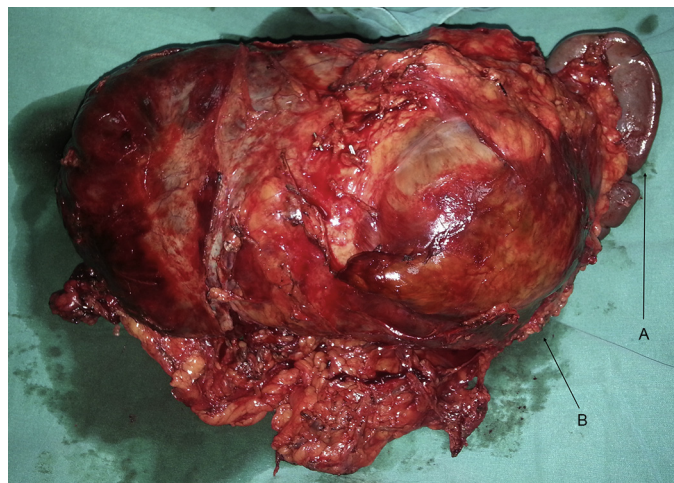


Fig. 2. The image of distal pancreatectomy and splenectomy specimen ((A) spleen and (B) distal pancreatic mass).

pancreas was made on fine-needle aspiration of the lesion guided by ultrasonography. The diameter of the lesion was increased probably a cystic hemorrhage after biopsy with a 25 cm at the widest diameter in the abdominal ultrasonography. Whole-body 18-fluorodeoxyglucose PET confirmed these findings, and surgery was planned by a multidisciplinary team. Preoperative diagnosis were pancreatic pseudocyst, cystadenoma or cystadenocarcinoma. At laparotomy, a cystic mass was found in the tail of the pancreas which was invaded to the transverse colon mesenterium and the spleen. We performed a frozen section from the mass and it was reported as benign intraoperatively. Distal pancreatectomy with splenectomy and transverse colon resection was performed. Mean operation time was 210 min. Mean blood loss was 800 cc

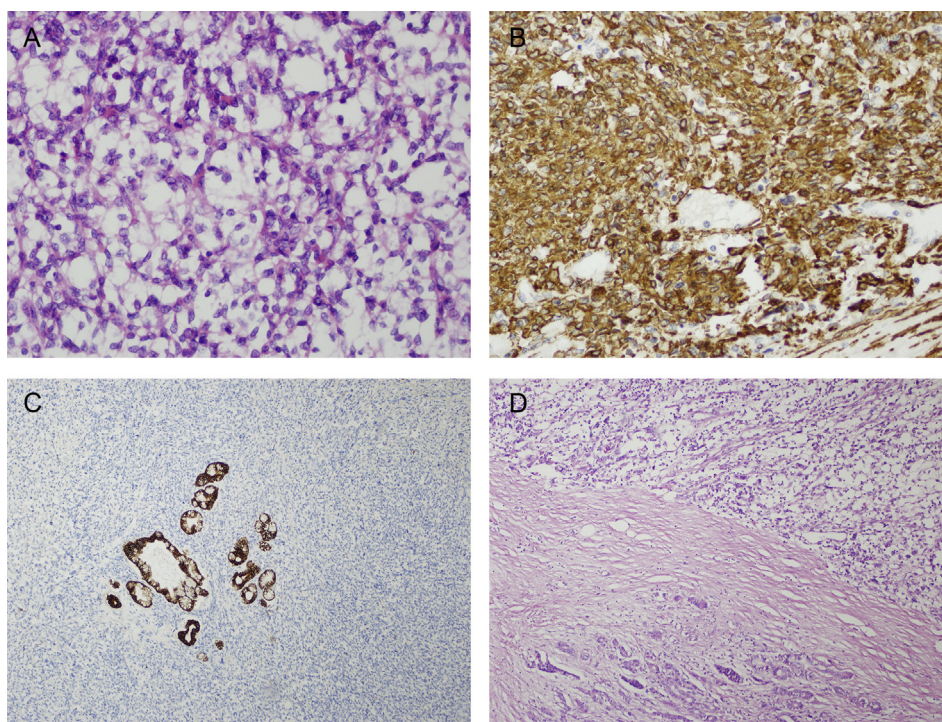


Fig. 3. The microscopic images of specimen with immunohistochemical staining. (A) Hematoxylin and Eosin (solid tumor consisting of plump round cells with mitotic activity), (B) smooth muscle actin (tumor cells positively reacting with SMA), (C) pancytokeratin (original pancreatic tissue entrapped by solid mesenchymal tumor) and (D) hematoxylin and eosin (tumor neighboring the pancreatic tissue).

and 3 unit of erythrocyte transfusion was performed intraoperatively. In recovery period, pancreatic leakage was occurred with a 100 cc flow daily. The patient was discharged 7 days after surgery and the pancreatic leakage finished spontaneously with a 2 month time. Gross examination of the surgical specimen revealed a 25 cm mass involving the tail of the pancreas which was partially cystic on cut section (Fig. 2). Histologically, the tumor was evaluated as poorly differentiated leiomyosarcoma. 11 mitototic figures per 50 high power fields were identified. The tissue lining the cystic cavity was composed of hemorrhagic areas. No necrotic areas were identified. Immunohistochemical staining demonstrated that the tumor cells were diffuse strong positive for smooth muscle actin, focally strong positive for desmin, positive for vimentin, negative for chromogranin, focally weak positive for Pancytokeratin (PANC-K) and S-100 (Fig. 3). Metastatic leiomyosarcoma was diagnosed histologically. She was treated neither chemotherapy nor radiation therapy. Six months postoperatively the patient is well without signs of recurrence.

3. Discussion

Metastatic lesions of the pancreas are uncommon and less than 2% of all pancreatic malignancies.^{1,3} The most common sites of the primary lesions were lung, kidney and gastrointestinal tract. However a few cases of leiomyosarcoma with metastases to the pancreas have been reported in the literature. Primary sites of leiomyosarcomas included the uterus, ovary, veins, spermatic cord, intestine, retroperitoneum and soft tissue.² Most patients have widespread disease on diagnosis of pancreatic metastases, whereas isolated metastases to the pancreas are rare.¹ The symptoms of pancreatic metastases are frequently nonspecific and generally include obstructive jaundice, pain and weight loss, which are very similar to the symptoms of primary pancreatic cancer.^{1,7} However, pancreatic metastases are frequently asymptomatic and detected during the follow-up.¹ The preoperative differential diagnosis between a metastatic lesion and a primary pancreatic neoplasm may be very difficult because of the similarity of signs and symptoms for both primary and secondary tumors.^{2,3} Accurate preoperative staging must be done to avoid major surgery in patients with widespread disease and 18-FDG-PET has proven to be useful for accurate staging.^{2,3} CT scan is usually performed to evaluate the extent of disease. Abdominal CT scans have revealed solid, cystic and heterogeneous images.^{6,7} Ultrasonography-guided fine-needle aspiration is helpful in the correct preoperative diagnosis, especially in patients with widespread disease.² Before deciding that the lesion in the pancreas was metastasis, primary leiomyosarcoma of the pancreas had to be ruled out. However, primary leiomyosarcoma of the pancreas is rare, and the differential diagnosis is very difficult because of the similarity between two.⁴ Also metastasis from tissues other than the uterus had to be ruled out secondly. So gastrointestinal tract, the soft tissues of the extremities and retroperitoneum must be investigated.⁴ Histologically, leiomyosarcoma of the pancreas contains interlacing spindle cells with varying degrees of atypia and pleomorphism which was originated from the walls of pancreatic blood vessels and pancreatic ducts. There is a strong relationship between number of mitoses per high-powered field and tumor aggressiveness. Other features like cellularity, degree of atypism and presence or absence of myofibrils have also been suggested for malign potential.⁷ Neither radiation therapy nor chemotherapy seems to be effective for leiomyosarcoma of the uterus. Giuntoli et al. studying 208 patients with leiomyosarcoma of the uterus and showed that adjuvant pelvic radiation therapy did not improve survival significantly. Omura et al. reported that there is no significant difference in the recurrence rate after adjuvant doxorubicin therapy.⁴ The surgical approach to the pancreatic

metastases must be aimed complete excision of the tumor with a wide negative margin of clear tissue and maximum preservation of pancreatic remnant if possible.^{5,7} Surgical options include pancreaticoduodenectomy, distal pancreatectomy, segmental resection and in some cases total pancreatectomy depends on the location of the lesion in the pancreas.⁵ Reported experience is limited and there is no established guideline regarding appropriate management of such lesions. When metastatic spread is limited to the pancreas, an aggressive surgery seems to increase survival and quality of life.^{1,2} The presence of extrapancreatic disease together with pancreatic metastases is not always a contraindication to resection, if it is technically possible.⁶ The survival rate after surgery is not clear and well-established because of the minority of cases but the curative surgery with wide margins seemed to be the best surgical option for these patients.^{1,2}

4. Conclusion

In the absence of widespread metastatic disease, aggressive surgical approach with negative margins must be aimed. Because it is associated with improved long-term survival and is the only hope for cure for these patients.^{1,6,7}

Consent

We obtained written and signed consent to publish a case report from the patient.

Conflict of interest

All of the authors (Safak Ozturk, Mutlu Unver, Burcin Kibar Ozturk, Osman Bozbıyık, Varlık Erol, Eyup Kebabcı, Mustafa Olmez, Nihat Zalluhoglu and Umit Bayol) certify that there is no relevant financial interest related to the material in the manuscript.

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Ethical approval

None.

Author contributions

Safak Ozturk MD concept and design, data collection, writing the paper; Mutlu Unver MD design and writing the paper; Burcin Kibar Ozturk MD radiological images and radiological diagnosis; Osman Bozbıyık MD data collection, Varlık Erol MD checking the language; Eyup Kebabcı MD data collection and concept of the study; Nihat Zalluhoglu MD data collection; Mustafa Olmez MD language checking; Umit Bayol MD pathological diagnosis and images.

Key learning points

We summarize the importance of R0 resection in pancreatic leiomyosarcoma metastasis. Because it is the only curative hope for these patients due to insufficient response to radiochemotherapy

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