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CASE REPORTS

Pancreaticoduodenectomy for metastasis of uterine leiomyosarcoma to the pancreas

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Abstract Metastasis of uterine leiomyosarcoma to the pancreas is rare. A 46-year-old woman was diagnosed with uterine leiomyosarcoma and underwent surgery. Thereafter, recurrences in the lung and subsequently in the pancreas were diagnosed. These lesions were resected and diagnosed as metastasis of uterine leiomyosarcoma. We report a rare case of uterine leiomyosarcoma with metastasis to the lung and pancreas, both of which were resected using aggressive surgery.

Keywords Metastasis · Pancreas · Uterine leiomyosarcoma · Pancreaticoduodenectomy

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Introduction

Leiomyosarcoma is a rare malignant tumour of smooth-muscle origin, originating most commonly from the female genital tract, gastrointestinal (GI) tract and soft tissues. The uterus is the most frequent site, and there the tumour has a high rate of recurrence and metastasis. The commonly reported sites of metastasis from leiomyosarcoma are the lung, kidney and liver; metastasis to the pancreas is extremely rare [1, 2]. Primary tumours that most commonly cause metastases to the pancreas are breast, lung, kidney and colon cancers or, more rarely, gastric cancer, melanoma and sarcoma Resection of metastatic tumour to the pancreas has occasionally been reported. Resection of pancreatic metastases was once uncommon; however, in recent years, improvement in morbidity and mortality rates after pancreaticoduodenectomy has been shown [3]. We present a rare case of metastasis to the pancreas of a uterine leiomyosarcoma.

Case report

In November 2003, a 46-year-old woman underwent an extended total hysterectomy, bilateral salpingo-oophorectomy and pelvic lymphadenectomy because of uterine epithelioid leiomyosarcoma of 13 cm with infiltration to the endocervix. In the extirpated uterus, there was a diffuse and fascicular proliferation of pleomorphic tumour cells with severe atypia. Tumour infiltrated the uterine serosa, and invasion to the lymphatic vessels was noted. The mitotic count was 20 per 10 high-power field. Immunohistochemical staining demonstrated that the tumour cells were positive for α-smooth-muscle actin. There was no metastasis to the ovaries or pelvic lymph nodes. The patient was diagnosed with uterine leiomyosarcoma stage I and underwent adjuvant radiation therapy because of tumour extension to the lower uterine segment. In early February 2004, a computed tomography (CT) scan of the chest and abdomen was obtained that demonstrated three small masses in left lung and chest lymphadenopathy. A positron emission tomography (PET)-CT scan was obtained that showed increased focal uptake of [^{18}F]-fluorodeoxyglucose (FDG) in the left

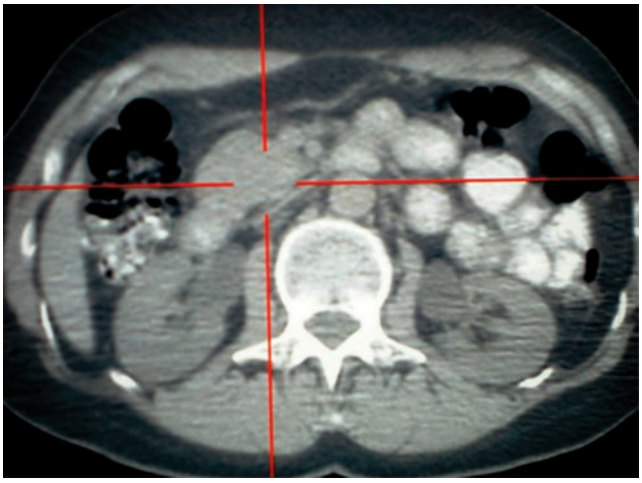

Fig. 1 Computed tomography scan of the abdomen shows a mass in the head of the pancreas

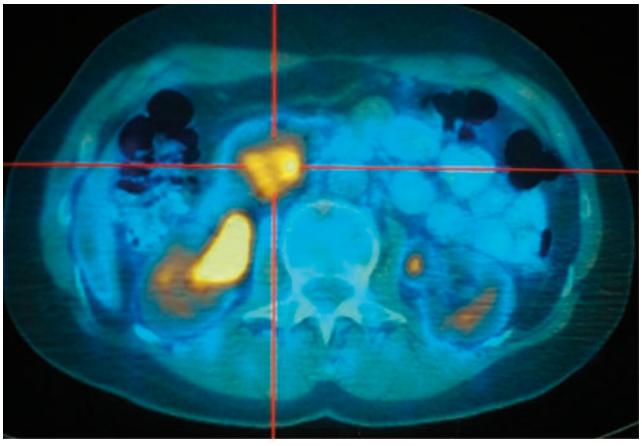

Fig. 2 Positron emission tomography shows increased focal uptake in the head of the pancreas

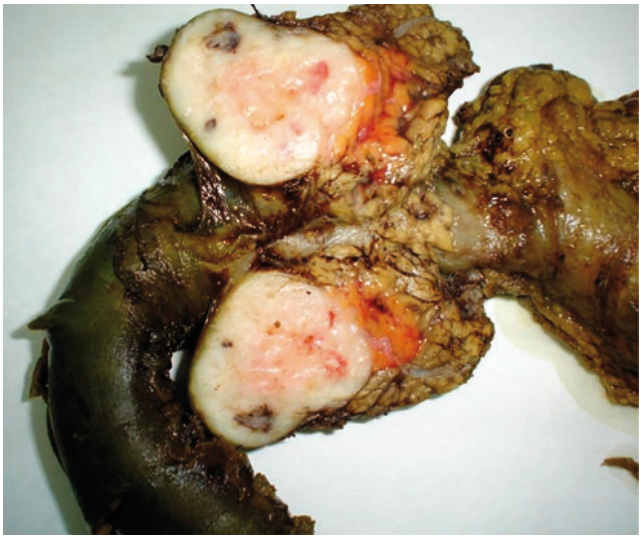

Fig. 3 Mass in the head of the pancreas

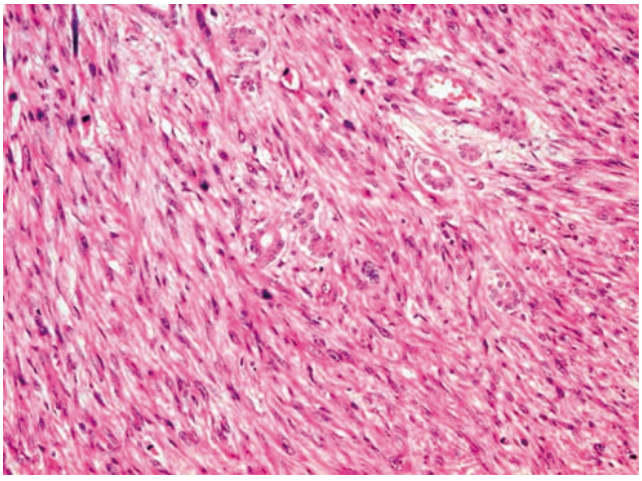

Fig. 4 Histology of the mass

lung (three masses) and no focal uptake in chest lymphadenopathy. In March 2004, the patient underwent chemotherapy with ifosfamide, Adriamycin and mesna. Thereafter, she was referred to the Department of Surgery when the lung metastases did not improve with chemotherapy. Given that the patient had no other evidence of metastatic disease, we offered her surgical segmentary resection and biopsy of the affected lymph nodes. The extirpated masses were approximately 1–2.5 cm, and the pathologic examination demonstrated a metastatic leiomyosarcoma. The mitotic ratio was as high as 30 per 10 high-power field. Immunohistochemical staining demonstrated that the tumour cells were positive for α-smooth-muscle actin. There was no lymph node invasion on biopsy. After the surgical resection, she underwent chemotherapy with gemcitabine.

At a routine follow-up visit in January 2008, a CT scan of the abdomen was obtained and demonstrated an approximately 4 cm mass in the head of the pancreas (Fig. 1). The patient felt asymptomatic. A PET-CT scan was obtained that showed increased focal uptake of [18F]-FDG in the head of the pancreas (Fig. 2), and endoscopic ultrasound (EUS) demonstrated a 4-cm heterogeneous lesion Fine-needle aspiration (FNA) was no available. Cancer antigen (CA) 19.9 and carcinoembryonic antigen (CEA) levels were within normal limits.

The patient was referred to our Department of Surgery a second time where she was offered surgical resection. In April 2008, after giving informed consent, she underwent a pancreaticoduodenectomy. The final pathologic examination demonstrated a metastatic leiomyosarcoma (Figs. 3 and 4). All margins were free of tumour. The mitotic ratio was 45 per 10 high-power field. Immunohistochemical staining demonstrated that the tumour cells were positive for α-smooth-muscle actin. The patient also had a biliary fistula, which resolved with conservative treatment. She has been seen in our hospital since discharged and was doing well when this report was written.

Discussion

We report a rare case of uterine leiomyosarcoma with metastasis to the pancreas. Uterine leiomyosarcoma accounts for 25–36% of uterine sarcoma and 1% of all uterine malignancies [1]. Metastatic lesions in the pancreas from this kind of tumour are uncommon. Primary tumours that most commonly cause metastases to the pancreas are breast,

lung, kidney (the most common) and colon cancers. At autopsy, the pancreas has been found to be a frequent site of metastasis in patients with these types of tumours (3–12%). However, the occurrence of solitary, potentially resectable, metastasis to the pancreas is less frequent (2%) [3–6].

Nakamura et al. reviewed autopsy records and pathology features of 103 cases of secondary pancreatic tumours from 690 cases of malignant tumours over a 10-year period and found the incidence was 15%. Gastric cancer was the most common primary tumour site (20%), followed by lung (18%) and extrahepatic bile duct (13%). They found that approximately half of the metastatic lesions were solitary [7]. Volkan et al reviewed 4955 adult autopsy cases and they found 190 cases with pancreatic tumors. 81 were metastases (43%). These were predominantly of epithelial origin, most commonly from lung (34), followed by GI tract (20) and kidney (4). Approximately one-fourth of them were in the head of the pancreas (13), body (1) and tail [6, 8].

Differential diagnosis of a primary pancreatic neoplasm from a metastatic malignancy may be very difficult. Symptoms and signs are similar for both primary and secondary tumours, and radiologic imaging may differentiate primary from secondary pancreatic lesions [3]. Good anamnesis, clinical aspects, FNA and findings of imaging techniques are necessary for accurate diagnosis and appropriate treatment of pancreatic metastasis [4]. Symptoms of pancreatic metastatic disease commonly include obstructive jaundice, pain, weight loss, new-onset diabetes and, more rarely, gastrointestinal bleeding and pancreatitis, as for primary pancreatic lesions. However, pancreatic metastases are frequently asymptomatic and detected during follow-up [8–11], as in our patient. Cytologic features along with clinical history can be used to develop a differential diagnosis and, more importantly, to help decide whether a lesion is benign or malignant. Cytopathologic features of the tumour cells combined with immunohistochemical evidence makes classification of this poorly differentiated neoplasm possible [12–14]. Imaging studies, such as CT scans and magnetic resonance imaging (MRI) may support the suspicion, especially if multiple tumours are noted. Highly vascular tumours, as indicated by contrast-enhanced CT, MRI or angiography, are more likely to be metastases than primary pancreatic cancers, which tend to be relatively hypovascular (except neuroendocrine tumours, which are also hypervascular). PET should be used routinely in the followed-up of patients with metastases as a highly sensitive, whole-body staging procedure [5, 10, 11].

Indications for pancreatic resections for metastasis have not been defined. Curative resection, surgical or endoscopic palliation or chemotherapy alone is then chosen according to the particular requirements of the clinical presentation and the expected outcome. The presence of limited extrapancreatic disease together with pancreatic metastasis is not always a contraindication to resection, if technically possible. In fact, resection of both pancreatic and limited extrapancreatic mass may be performed with low risk. However, in the absence of widespread metastatic disease, aggressive resection seems to be associated with improved long-term survival [3, 10, 11]. Pancreaticoduodenectomy can be performed safely, representing a suitable option for resection in patients with isolated pancreatic metastases in the absence of widely metastatic disease. The best indications are solitary metastases from renal-cell carcinoma, sarcoma, colon carcinoma and neuroendocrine tumours [3, 4, 9]. Radical surgical resection has been proven to be associated with improved survival and a better quality of life. In cases of unresectable disease, surgical or endoscopic palliation in association with chemotherapy can improve the quality of life and, in selected but rare cases, may achieve prolonged survival [6].

Conclusion

Radical surgical resection has been proven to be associated with improved survival and a better quality of life. In cases of unresectable disease, surgical or endoscopic palliation in association with chemotherapy can improve the quality of life and, in selected but rare cases, may achieve prolonged survival. In our case, the patient underwent pancreatoduodenectomy because of leiomyosarcoma metastasis to the pancreas. With this surgery, to date, the patient has had no further recurrence; careful and strict follow-up is needed.

Conflict of interest The authors declare that they have no conflict of interest relating to the publication of this manuscript.

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