症例報告 A huge abdominal leiomyosarcoma 6 years after an initial hysterectomy for a leiomyoma : Report of a case with a diagnostic dilemma

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A huge abdominal leiomyosarcoma 6 years after an initial hysterectomy for a leiomyoma: Report of a case with a diagnostic dilemma

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ABSTRACT

A case of a huge abdominal leiomyosarcoma 6 years following an initial hysterectomy for a leiomyoma in a 64 year old female, is reported. The patient was referred to the Ryukyu University Hospital with a diagnosis of an abdominal tumor on June 7, 1994. She had a history of hysterectomy at age 58 for a leiomyoma. Abdominal CT scans demonstrated a large, conglomerate tumor with metastatic masses. Exploratory laparotomy revealed two large tumors with extensive peritoneal dissemination. To establish the exact nature of this abdominal tumor with pathologic examination and intend tumor reduction, the largest tumor of the omentum 24×20 cm in size was resected. The pathology report was metastatic leiomyosarcoma. This condition led us to review the original uterine slides, which showed similarities with this abdominal tumor, we concluded that the abdominal tumor was a recurrent leiomyosarcoma of uterine leiomyosarcoma. In conclusion, patients with problematic uterine smooth muscle tumors should be observed closely over a long period. Ryukyu Med. J., 15 (3) 143-146 1995

Key words: recurrent leiomyosarcoma, hysterectomy, uterine leiomyoma

INTRODUCTION

So-called benign metastasizing leiomyoma is a rare condition that usually appears in the organs of middle-aged women many years after an initial hysterectomy for leiomyoma14. However, some cases, at least in retrospect, may be described as low grade leiomyosarcomas14. In clinical practice, this condition can present a diagnostic problem to general surgeons, gynecologists, and pathologists. We present a case in which a huge abdominal metastasis was diagnosed 6 years after hysterectomy for a leiomyoma with histological features of some degree of cytologic atypia and mitotic activity.

CASE REPORT

A 64-year-old female presented with mild pain and some distention of the lower abdomen in June 1994 at a local hospital. An abdominal CT scan demonstrated a conglomerate, large mass probably arising from the omentum with some metastasis to the upper abdominal cavity. On June 7, 1994 the patient was referred to the Ryukyu University Hospital for surgical treatment. She had a history of brain vascular disease, which caused her to react aggressively at age 57, and hysterectomy with partial ileal resection at age 58 for a leiomyoma.

On admission, the patient appeared physically healthy, but had an aggressive nature. The physical examination revealed alarge, soft, movable mass palpable in the lower abdomen with a lower mid-line surgical scar. The laboratory findings showed 10.7 g/dl of hemoglobin, 886 IU/L of LDH, normal ranges of liver function tests and normal titers of tumor markers (AFP, CEA, CA 19-9). Abdominal ultrasonography (US) disclosed a large, hyperechoic mass with internal heterogenous echoic patterns in the middle to lower abdomen. Abdominal CT scan demonstrated a huge, well-delineated tumor 25×20 cm in size occupy in the space from the renal hilus to the rectovesical excavation. Its internal density appeared heterogeneous. Another mass 6×7 cm in size was found below the liver with internal fluid-fluid level formation. These diagnostic modalities indicated that this tumor arose from the omentum and was a kind of sarcoma (Fig.1).

On June 20, exploratory laparotomy was performed. The abdominal cavity contained a small amount of bloody ascites. Two large tumors with extensive peritoneal dissemination were observed. The one in the lower abdomen was thought to arise from the omentum and to be primary tumor, while the other in the right upper quadrant of the abdomen was considered as a metastatic tumor. However, it was impossible to establish the exact nature of a given tumor without pathologic examination of the resected tumors. Only the huge tumor was resected to establish its exact nature. Further surgical treatment was not consider-
ed in view of the extensive and multiple distribution of the tumor in this patient.

The resected tumor was $24 \times 20$ cm in size. It was soft, fragile and hemorrhagic in nature. A section showed extensive hemorrhagic necrosis with clotted blood (Fig. 2). Microscopic studies of the resected tumor showed interlacing clusters of spindle cells with eosinophilic cytoplasm and irregular, bland nuclei. Cytologic atypia and mitotic figure were identified. The mitotic count was 5 mitotic figures per 10 high power microscopic fields (5 MF/10 HPF) of the most cellular area (Fig. 3, bottom).

Immunohistochemically, the tumor was positive for Vimentin, and stained red with Azan staining. Based on these findings, this tumor was diagnosed to be leiomyosarcoma, metastatic.

A review of the original leiomyoma slides of the uterus and the ileum removed 6 years earlier, showed similar histological abnormalities. There were some cytologic atypia and mitosis without evidence of coagulative tumor cell necrosis compatible with a leiomyosarcoma. The ileal tumor lied adjacent to the uterus. The uterine slide had 2 MF/10 HPF, and the mitotic count of ileal slide was 4 MF/10 HPF.

We found good interobserver concordance in the interpretation of the original slides. In the result, the original uterine tumor was evaluated to be low grade leiomyosarcoma. Subsequently, our final diagnosis in this patient was recurrent abdominal leiomyosarcoma secondary to primary uterine leiomyosarcoma. The patient started with adjuvant chemotherapy (Epirubicinhydrochloride, Cisplatin and Etoposide) after reduction surgery. On the seventh post operative day, we injected Epirubicin hydrochloride of 30mg intravenously, On the eighth day, We injected Cisplatin of 50mg by dropping injection From the fourth to sixth postoperative day, Etoposide of 160mg was injected intravenously. After completion of chemotherapy, abdominal CT scan demonstrated response to residual abdominale tumors. Six months later, on January, 1995, she was again hospitalized for adjuvant chemotherapy for growing residual abdominal leiomyosarcoma. By CT scan, The residual tumor size had increased from $5 \times 5$ cm to $12 \times 14$ cm in tumor diameter.

**DISCUSSION**

Because this patient, s uterine tumor was initially reported to
be a leiomyoma, the abdominal tumor on her admission led us to suspect a primary malignant tumor arising from the omentum or the retroperitoneum. However, exploratory laparotomy revealed recurrent or metastatic abdominal leiomyosarcoma of an unknown origin.

The clinical issue in evaluating any group of neoplasms of a particular differentiated type is to precisely determine which morphologic features reliably predict clinical relapse and clinical outcome. For uterine smooth muscle neoplasms with histologic features that are not unequivocally benign or unequivocally malignant, this evaluation has been a continuing challenge. For instance, so-called benign, metastasizing leiomyomas have been described as an uncommon condition usually seen many years following hysterectomy for a benign leiomyoma. This puzzling condition has been attributed to vascular invasion or intravascular leiomyomas in the uterus, with subsequent hematologic dissemination into the distant organs. The alternative thought is that these tumors are essentially low grade leiomyosarcoma and capable of recurrence or metastasis. On the histologic examination, some of the cases show low grade malignant features, while others reveal apparent benign features.

On review of the slides, our case showed the uterine tumor to have malignant features, of which misinterpretation led us to preoperative diagnostic error. It is widely recognized that lesions with fewer than ten mitoses in ten high-power fields prove benign on follow-up, whereas tumors with ten or more mitotic figures in ten high-power fields metastasize. Mitotic index itself is not standardized and often unrepeatable because of various factors, such as the thickness of the sections, area examined, lack of an acceptable uniform morphologic criterion for mitosis and observer's subjective variations. Another reason for the poor performance of the mitotic index may be the presence of steroid receptors in the uterine smooth muscle cells. Accordingly, hormone levels can be reflected in an increased mitotic index in both normal uterine muscle and leiomyomas.

Other investigators have focused on the importance of cytologic atypia or more recently combination of three histopathologic features (mitotic index, cytologic atypia and coagulative tumor cell necrosis) in establishing criteria for leiomyosarcoma. Even marked cytologic atypia when coupled with a low mitotic index is often not associated with a clinically malignant course. This fact degrades the performance of cytologic atypia as a predictor of clinical outcome. Combination of three morphologic features has been much recently described best to predict clinical outcome. In our case, the initial uterine tumor showed mild to moderate cytologic atypia and mitotic index, but no evidence of coagulative tumor cell necrosis.

In conclusion, although the risk of recurrence of uterine smooth muscle neoplasms many years following hysterectomy for a leiomyoma may be uncommon, continuing follow-up seems to be justified in patients with histopathologically problematic uterine smooth muscle neoplasms.

REFERENCE

1) Lipton, J.H., Fong, T.C., and Burgess, K.R.: Miliary pattern as presentation of leiomyomatosis of the