Recurrent Uterine Leiomyosarcoma Implanted in a Laparotomy Scar

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Uterine leiomyosarcoma, one of the most malignant groups of uterine tumors, constitutes 0.1–2% of all uterine malignancies. The pattern of tumor spread is commonly to the liver or lung via the blood stream. The treatment of most patients with stage I and II uterine leiomyosarcomas should include at least total hysterectomy and bilateral salpingo-oophorectomy. There is no firm evidence that adjuvant chemotherapy or radiation therapy is beneficial for these patients. However, recurrences develop in more than half of the cases of uterine leiomyosarcoma, even when the disease is apparently localized at the time of treatment.

The most common sites of recurrence are the abdomen and lungs, and less than 10% of cases showed recurrence in the pelvic cavity. However, recurrences in the laparotomy wound are extremely rare and, to our knowledge, only one case has been previously reported [1]. It is still not known how neoplastic cells can implant and grow in an abdominal scar without other concomitant metastases.

We present a patient with uterine leiomyosarcoma who developed a recurrence in her abdominal wall incision and review the reports in the literature about potential etiologies as well as the mechanism.

A 43-year-old woman, gravida 3, para 2, presented to our gynecologic oncology clinic in February 2004, complaining of self-palpable abdominal mass associated with intermittent lower abdominal pain for more than 1 year. On pelvic examination, an enlarged uterus, the size of which was similar to that at 12 weeks' gestation, was noted, and transvaginal sonography revealed a huge uterine tumor of about 12 cm in diameter with increased vascularity. A laparotomy was performed through a Pfannenstiel incision. Findings included a huge uterine tumor measuring 12 × 9.5 × 8 cm with central necrosis, and there was no obvious disease involving the bilateral ovaries, lymph nodes or omentum. The patient underwent total abdominal hysterectomy, because an atypical leiomyoma was disclosed from intraoperative frozen section. The final pathologic evaluation was reported to be uterine leiomyosarcoma with mitotic count higher than 10 per 10 high-power fields. There was no evidence of uterine serosal involvement. No adjuvant treatment, such as chemotherapy or radiotherapy, was arranged after the surgery.

The patient was free of disease until 2 years after the operation, when a solitary mass was noted at the laparotomy scar. On examination, she had a 4 × 4 cm hard mass with restricted mobility in the right side of the Pfannenstiel scar. Pelvic examination and serum tumor markers were within reference ranges. Ultrasonography of the abdomen showed one subcutaneous cystic mass that was 4 cm in diameter near the laparotomy scar. The abdominal computed tomography scan revealed a 4-cm, well-enhanced mass at the right lower anterior abdominal wall, and there was no evidence of intra-abdominal or distant disease (Figure 1). Under the impression of recurrent leiomyosarcoma, local excision of the mass

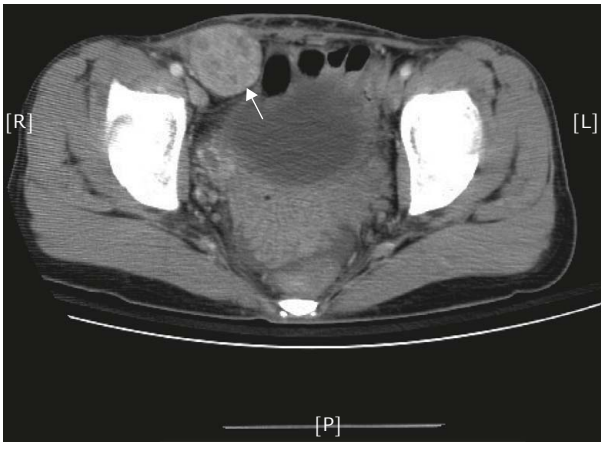

Figure 1. Abdominal computed tomography showing a 4-cm, well-enhanced mass (arrow) with a thick tail at the right lower anterior abdominal wall, separating from the urinary bladder.

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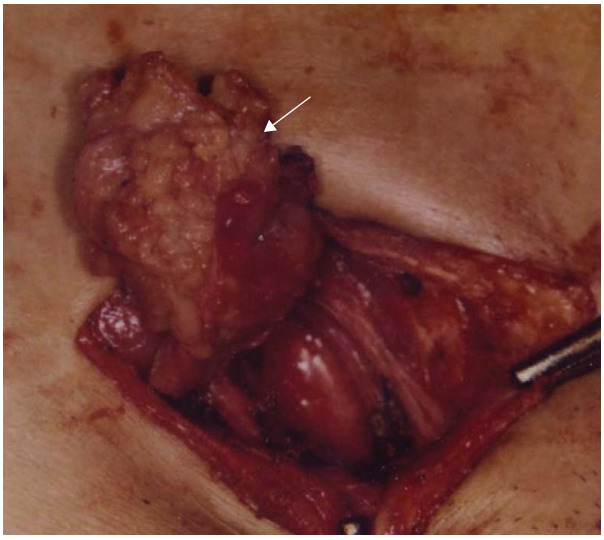

Figure 2. A 4×4 cm mass (arrow) over the right side of the previous operation area, without entering into the pelvic cavity.

through the previous Pfannenstiel incision was undertaken (Figure 2). Perioperatively, the mass was found to be hard, of 4 cm in size, and was located inferior to the fascia of the right side of the previous laparotomy wound. On microscopy, the tumor cell was high-grade leiomyosarcoma and very similar to that of the primary tumor. Additional immunohistochemical study also showed positive expression for desmin and smooth muscle actin, consistent with smooth muscle differentiation. However, the surgical margin was not clear of tumor cells. Postoperatively, after the wound healing, this patient received adjuvant radiotherapy to the whole pelvis with total dose of 5,040 cGy in 28 fractions, five times a week.

Unfortunately, a recurrent mass, 5.7 cm in diameter, was noted over the right side of the pelvic cavity 4 months after radiotherapy, while the laparotomy wound was cancer free. No metastasis of the abdominal lymph nodes was detected. Subsequently, the patient sought a second opinion at another hospital and underwent a second laparotomy for a tumor-debulking surgery. The final pathologic evaluation of the tumor mass was reported to be consistent with recurrent leiomyosarcoma. Postoperatively, she received adjuvant chemotherapy with dacarbazine and epirubicin, and remained disease-free for 9 months after the tumor excision.

Recurrences develop in more than half of the cases of uterine leiomyosarcoma [1]. The most common sites of recurrence are the pelvis, lungs and liver. In contrast, metastasis in the abdominal wall incision line has rarely been described [2]. To our knowledge, this is the second reported case of recurrent leiomyosarcoma implanted in an abdominal scar. Cancer recurrence at the site of a surgical wound is a well-documented but relatively rare event, especially in laparotomy scars. However, laparoscopic port-site metastases in patients with malignancies have been reported, including in those with carcinoma of the pancreas, esophagus, stomach, liver and colon, as well as gynecologic malignancies [3]. In order to explain the development of laparoscopic port-site metastasis, a number of etiologic factors have been proposed, such as "pneumoperitoneum" and "the chimney effect" [4,5]. Although these concepts remain controversial, some researchers claimed that the surgical technique actually resulted in increased tumor growth and a higher incidence of port-site or abdominal wall metastases, compared with gasless laparoscopy. Other potential etiologies that have been discussed in the literature include local immune system, surgical technique, and direct contamination of the trocar site with viable tumor cells [6].

However, how the cancer cells implant themselves in the laparotomy wound and grow *in situ* is still not known. Furthermore, the presentation of a solitary recurrent tumor mass at a surgical incision wound without other concomitant metastatic sites raises the question of the route of spread. Positive peritoneal cytology is one of the factors which contributes to recurrence and poor prognosis [7]. However, only a few reports show an increased risk of recurrence at the site of surgical wound if positive cytology is proved, despite the common presence of cancer cells intraperitoneally [8]. Some authors believed that malignant tumor cells implant in the laparotomy wound at the time of surgery because of tumor cell spillage during manipulation, and then grow slowly, as a mechanism for skin incision metastases. Therefore, the importance of a sterile operative field to avoid cancer cell contamination has been emphasized for a long time [9,10]. In addition, Kotwall et al [11] reported an as-yet-unknown interplay between the host, tumor and treatment factors that determines the viability of implanted tumor cells. In uterine tumors, tumor grade is also one of the most important factors in local, regional and distant recurrence. High-grade tumors with poorly differentiated cells have the greatest propensity for implantation at the incision site, as in our patient [1].

Management methods of recurrent uterine leiomyosarcoma at the site of a surgical wound have been reported, but no strict guidelines are available in the literature owing to the scanty number of cases reported. Nair et al [1] presented a woman with high-grade spindle cell sarcoma of the uterus, who underwent radical surgical resection of isolated scar metastasis. Then, the patient received postoperative adjuvant chemotherapy followed by external radiation. No evidence of recurrent tumor was noted during the 18 months of follow-up. Although radical surgical excision with postoperative

adjuvant radiotherapy or chemotherapy are both suggested, especially when the resection margins are not clear [12], the outcomes remain uncertain. We treated our patient with local excision followed by external beam radiation therapy. Unfortunately, she was found as having recurrent mass in the pelvic cavity, other than at the laparotomy scar, 4 months after radiation. Whether giving chemotherapy soon after surgical resection of scar tumor would have benefited this patient still needs further investigation.

In conclusion, laparotomy scar recurrence following treatment for gynecologic cancer without other concomitant metastases is rare, and the route of spread remains uncertain. In our case, a possible explanation is the direct tumor seeding in a previous laparotomy scar due to cell spillage during manipulation at the time of surgery. To avoid cancer recurrence at the site of surgical wounds, we emphasize the importance of a sterile operative field and irrigation of the pelvic cavity as much as possible when malignancy is suggested.

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