Excision of abdominal wall recurrence of uterine leiomyosarcoma: A case report

Sahana Punneshetty¹, Rachel G Chandy², Geley Ete³, Suchita Chase⁴, Anjana Joel⁵, Abraham Peedicayil²

From ¹Senior Resident, ²Professor, Department of Gynaecologic Oncology, ³Associate Professor, Department of Plastic Surgery, ⁴Professor, Department of General Surgery, ⁵Assistant Professor, Department of Medical Oncology, CMC, Vellore, Tamil Nadu, India

ABSTRACT

The literature on management of recurrent/metastatic uterine leiomyosarcomas is limited and metastasis to the abdominal wall is very rare. We operated on a 46-year-old woman who had a total abdominal hysterectomy elsewhere for a uterine tumor and presented with recurrence to the anterior abdominal wall, 9 months after the primary surgery. The challenge after complete excision of the tumor was to reconstruct the abdominal wall. We were able to close the defect with a dual mesh and a fasciocutaneous flap from the anterolateral thigh. She received adjuvant chemotherapy with gemcitabine and docetaxel. At the completion of chemotherapy, she was well and apparently disease free. She recurred in the left inguinal node after 9 months but responded to oral pazopanib followed by chemotherapy. This case report highlights the rare possibility of abdominal wall metastasis after initial surgery for leiomyosarcoma. It shows that good quality of life can be achieved by a multidisciplinary treatment of abdominal wall metastasis of uterine leiomyosarcoma.

Key words: Abdominal metastasis, Case report, Reconstruction, Uterine leiomyosarcoma

CASE REPORT

A 46-year-old unmarried woman presented to us with a non-healing ulcerative abdominal wall mass of 3 months duration. She had hypertension on telmisartan 40 mg daily. Her father had cancer of the lung.

She had had a total abdominal hysterectomy done elsewhere, a year earlier, for heavy menstrual bleeding. Histopathology was atypical leiomyoma with myxoid degeneration. No further therapy was given. After surgery, the patient was well for about 8 months and then noticed an abdominal tingling sensation and non-healing ulcers at the surgical scar for which she underwent incision and drainage multiple times. She then noticed a swelling at the previous scar. The timeline is shown in Table 1. In view of a non-healing ulcer, she underwent a biopsy of the lesion which was reported as a poorly differentiated malignant tumor positive for vimentin, SMA, and Ki 67, and was referred to a tertiary center for further management.

On admission, general examination showed that there was no pallor or lymphadenopathy. Her pulse rate was 104/min, blood pressure was 110/60 mmHg, and functional status was Eastern Cooperative Oncology Group 1. She had two large (9 cm×7 cm and 7 cm×5 cm) adjacent abdominal wall lumps (Fig. 1a) with surface ulcers. There were two large lobulated heterogeneously enhancing soft-tissue lesions with large areas of central necrosis in the lower anterior abdominal wall, centered in the subcutaneous and muscular plane with posterior extension into the pre-peritoneal space. Vulva and vagina were normal. Rectal examination was also normal.

Her biopsy slides were reviewed by our pathologist. Her positron emission tomography and computed tomography scan (Fig. 1b) showed that lesions were confined to the abdominal wall. A primary abdominal wall tumor was considered less likely than a recurrence from uterine sarcoma. The slides were reviewed at our hospital and were reported as high-grade sarcoma. The clinicopathologic features.

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Correspondence to: Abraham Peedicayil, Department of Gynaecologic Oncology, CMC Hospital, Vellore - 632 004, Tamil Nadu, India. E-mail: apeedicayil@yahoo.com

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DISCUSSION

LMSs are the most common sarcomas accounting for 1–2% of all uterine malignancies with an incidence of 0.3–0.4/100,000 women per year [2]. The most common age of presentation is 45–55 years with a median age of 52 years. The common symptoms include abnormal vaginal bleeding (56%), palpable pelvic mass (54%), and pelvic pain (22%). They are more likely to have early hematogenous spread. Most of the tumors are aggressive with a poor prognosis and high local recurrence rate.

They usually arise de novo from uterine smooth muscle. Pelvic radiation [4] and the use of combined hormone replacement therapy in postmenopausal women for 5 years or longer are also associated with an increased risk [5]. Uterine curetting is positive in only 10–20% [6]. The tumors are usually larger than 10 cm in size. The cut surface is typically soft, bulging, fleshy, necrotic, and hemorrhagic with irregular margins.

Uterine LMSs consist of highly cellular, spindle-shaped smooth muscle cells, mitotic figures, and coagulative necrosis. The histological subtypes include spindled, epithelioid, and myxoid types. The spindled variant accounts for most cases with a very high mitotic index [7]. Tumors are graded into low grade and high grade depending on cellular atypia, mitotic index, and the presence of necrosis which are the main prognostic factors for...
recurrence. The most common immunohistochemistry markers are desmin, h-caldesmon, smooth muscle actin, and histone deacetylase. They also express estrogen receptors, progesterone receptors, and androgen receptors. LMSs express higher levels of Ki-67 than leiomyomas. The genetic profile suggests genomic instability which is a hallmark of uterine sarcomas. The presence of smooth muscle actin is nearly uniform and desmin positivity is usual. This along with the lack of KIT expression separates leiomyosarcoma from the gastrointestinal stromal tumors, an important problem in abdominal soft tissues [8].

Surgery is the mainstay of treatment and includes total abdominal hysterectomy with bilateral salpingo-oophorectomy and debulking of any tumor outside the uterus including enlarged nodes. Stage is the most powerful prognostic factor. Spindle cell LMSs recur within 2 years compared to other variants which recur late. There is sparse literature on the management of recurrent uterine leiomyosarcoma. Studies have demonstrated that the patients with recurrent uterine sarcomas who underwent secondary cytoreductive surgery had longer overall survival and disease-free intervals following the initial relapse when compared with those treated with chemotherapy or radiotherapy alone. Surgery for extrapulmonary recurrences of uterine sarcomas is limited [9].

Chemotherapy with or without palliative radiation therapy is generally recommended for patients with Stages II and III disease, incompletely resected, or metastatic disease [10]. The chemotherapeutic agents used for recurrent disease are fixed-dose gemcitabine and docetaxel, single-agent doxorubicin, paclitaxel, trabectedin, eribulin, megestrol acetate, aromatase inhibitors, and pazopanib. Adjuvant radiation therapy is considered for patients with high recurrence risk as it has shown to reduce the pelvic relapse rate by 50%. Cutaneous metastasis of uterine leiomyosarcoma has been reported earlier [11]. Secondary cytoreduction may be considered, as neither chemotherapy nor radiation improved outcomes in recurrent disease [12]. Secondary cytoreduction is associated with prolonged survival in patients with isolated disease recurrence and long progression-free interval from the primary disease [9,13].

CONCLUSION

The management of recurrent LMSs and advanced-stage disease depends on resectability. Surgery and metastasectomy, in properly selected patients, may improve quality of life and survival. Multimodality treatment and palliation should also be considered.

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REFERENCES


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