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Cervical Leiomyosarcoma: A Case Report

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Abstract

Introduction: Sarcoma of the cervix is uncommon and its prevalence is about 1% of malignancies of the cervix. One type of sarcoma is leiomyosarcoma and the location of this tumor in cervix is very rare. Although uterine sarcoma can involve the cervix and lead to misdiagnosis of cervical leiomyosarcoma, the location of the bulk of the tumor in uterine cervix (not isthmus) for diagnosis of cervical leiomyosarcoma is mandatory. The origin of sarcoma is from mesodermal tissue. Due to the rarity of cervical leiomyosarcoma and response to debulking surgery after recurrence, report of this case is interesting.
Case Presentation: We hereby report on a 34-year-old unmarried patient, who was a known case of leiomyosarcoma of uterine cervix that was referred with recurrence of tumor in parietal peritoneum, retropobic retzius space and around the bladder and ureter and sigmoid colon. The patient was treated with debulking surgery of the tumor and chemotherapy.
Conclusions: Sarcoma as a genital tract malignancy is very rare and the location of sarcoma at pediatric age is in the vagina and at middle age is in the cervix and at postmenopausal women in the uterine corpus. Cervical sarcoma as a rare tumor of the cervix arises from stromal tissue. Due to hematogenous spread of sarcoma, metastasis in lungs is seen and recurrence of this tumor is not uncommon. In this article, recurrence of a known case with leiomyosarcoma of uterine cervix in previous hysterectomy that responded to debulking surgery with removal of metastatic lesions in peritoneal and retroperitoneal spaces and adjuvant therapy with standard chemotherapy drugs and monoclonal antibody therapy is reported. Due to non-hormonal dependence of sarcoma to hormonal secretion from the ovaries and benefit of the ovaries in young women for protection of bone health and cardiovascular system, oophorectomy is a challenging idea and an issue of debate, thus primary surgery, in this case oophorectomy, was not done.

Keywords: Cervical Sarcoma, Leiomyosarcoma, Uterine Cervix, Adjuvant Therapy

1. Introduction

Prevalence of sarcoma of the cervix is about 1% and leiomyosarcoma of the cervix is very rare (1, 2). Primary leiomyosarcoma of the uterine cervix is extremely rare (3). In 2008, review of literature showed rare incidence of leiomyosarcoma of the cervix, where 21 cases have been reported (3). Leiomyosarcoma of the cervix occurred in premenopausal women, but leiomyosarcoma of the body of uterus occurred in postmenopausal women. The most common symptoms of patients includes vaginal bleeding and mass in the cervix (4).

This case report is concerned with the recurrence of leiomyosarcoma of the cervix in a 34-year-old female, and probably one of the first cases of cervical leiomyosarcoma in a young woman. Our case was unique, since it was recurrence of the disease in a young woman, who was treated with combined modality of the treatment. On the other hand, after two years of close follow-up, the disease responded to the treatment and the patient survived.

2. Case Presentation

A 34-year-old unmarried female, who was known case of leiomyosarcoma of uterine cervix, referred to Firoozgar hospital. One and a half years before her referral, after a six-month course of heavy menstrual bleeding, pelvic pain and urinary retention, the patient underwent laparotomy. Magnetic resonance imaging (MRI) investigations before laparotomy had revealed a 14 cm enhancing mass lesion in the anatomic location of the cervix with remarkable protrusion to the vaginal cuff and remarkable pressure effect on adjacent organs.

During the operation, the surgeon encountered a large necrotizing mass in the cervix and frozen section demonstrated high-grade leiomyosarcoma, which was compatible with permanent report, hence, total abdominal hysterectomy was done. The patient then underwent three courses of chemotherapy with taxotere and gemcitabine.

Post-operative follow-up evaluations revealed a 5 mm lung nodule and sub pleural nodule, enhanced foci upon

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the sigmoid wall, lower rectus sheath and bed of the hysterectomy six months after the operation. Evaluation with a cystoscope also demonstrated transposition of right ureteral orifice due to mass effect of an extra luminal lesion. The same external pressure was obvious in colonoscopy.

The patient referred to our center with an impression of recurrence of leiomyosarcoma. On physical examination, the abdomen was soft and two soft and tender masses measuring about 4.5 × 2 cm were palpable in right and left portion of the previous skin scar. Rectovaginal exam was normal and vaginal examination was not possible because the patient was virgin. Figure 1 shows metastatic lesions in the abdominal wall.

After a second course of chemotherapy with 5-(3,3-dimethyl-1-triazeno)-imidazole-4-carboxamide (DTIC) and adriamycin, the patient underwent the debulking operation and metastatic lesions of sigmoid serous, rectus sheath and bladder serous were resected. One metastatic lesion measuring 5 × 6 cm adhesive to parietal peritoneum was resected. After ureterolysis, right salpingo-oophorectomy was done because of gross involvement, but the left ovary was spared. Permanent pathology confirmed recurrence of high-grade pleomorphic leiomyosarcoma in abdominal wall masses, right ovary and fallopian tube, subfacial masses, peritoneal mass and mesosigmoid masses (Figure 2). The patient received two other courses of DTIC and adriamycin. The disease was stable according to the response evaluation criteria in solid tumors (RECIST). Six-month later, follow-up CT scan revealed pulmonary nodule and recurrence of disease in bladder and sigmoid. Therefore, the patient received four more courses of chemotherapy with thalidomide and methazolamide and radiotherapy. However, no response was obvious and the patient received sunitinib. The disease was stable for seven months, but imaging investigations revealed pulmonary nodule and the patient received sorafenib.

3. Discussion

Leiomyosarcoma of the cervix is very rare and occurs mostly in premenopausal women (5). Diagnosis before surgery is a rare occurrence (6). In leiomyosarcoma of the uterine corpus, involvement of the cervix is not uncommon, so for definite diagnosis of primary cervical leiomyosarcoma, the bulk of the tumor should be on the cervix but not the isthmus (1). These tumors occur generally in perimenopausal period, but one case with leiomyosarcoma of the cervix was reported in a teenage (7).

In a 2001 article, Gotoh et al. reported epithelioid leiomyosarcoma of the cervix in a 72-year-old woman (8). The patient underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy. Our patient had undergone total abdominal hysterectomy followed by chemotherapy. Recurrence of the disease occurred. Debulking surgery was performed and the patient received radiotherapy and chemotherapy.

In 2013, Doshi et al. reported leiomyosarcoma of the cervix in a 54-year-old woman, who had undergone Wertheim hysterectomy without radiotherapy or chemotherapy and the patient was disease-free during a six-month follow-up (5).

In 2003, Irvin et al. reported leiomyosarcoma of the cervix in a 47-year-old woman, who underwent modified radical hysterectomy and bilateral salpingo-oophorectomy (9).

In 2015, Mehra et al. presented a 38-year-old woman, who was cured with standard surgery and radiotherapy with chemotherapy four weeks post total abdominal hysterectomy with bilateral salpingo-oophorectomy (6).

In 2013, Dhull et al. reported leiomyosarcoma of the cervix in a 34-year-old woman, who had undergone simple hysterectomy but later histopathology revealed leiomyosarcoma of the cervix hence bilateral salpingo-oophorectomy was not performed. Post-operative chemotherapy was performed (10).

In 2008, Sahu et al. reported leiomyosarcoma of the cervix in a 25-year-old woman, who underwent neoadjuvant chemotherapy and then total abdominal hysterectomy and bilateral salpingo-oophorectomy and radiotherapy was done (3).

In general, total abdominal hysterectomy and bilateral salpingo-oophorectomy represents the standard treatment for cervical leiomyosarcoma. However, in low-grade tumor and premenopausal patients, there is debate about oophorectomy.

In the present case, the patient was unique, since there was recurrence of the disease in a young woman, who was treated with combined modality of treatment. On the other hand, after two years of close follow-up, the disease responded to the treatment and the patient survived.

Footnotes

Authors' Contribution: Sahereh Arabian, drafting the manuscript; Shirin Haghighi, chemotherapist; Ladan Haghighi, the surgeon for primary hysterectomy.

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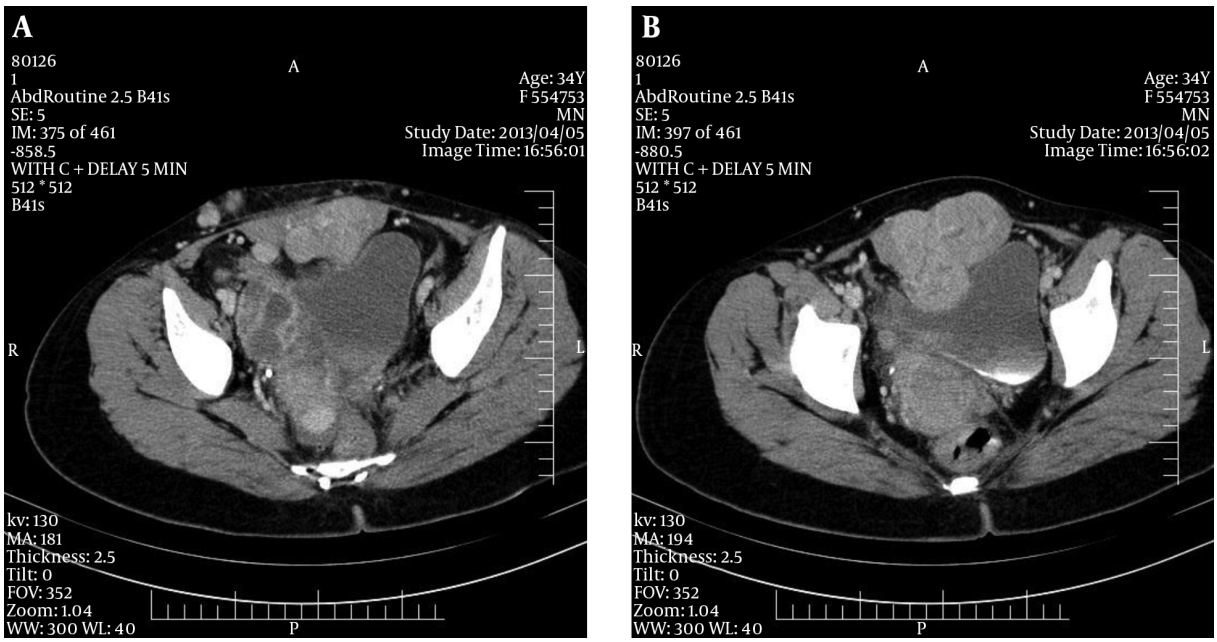


Figure 1. Axial View of Metastatic Lesions in Abdominal Wall

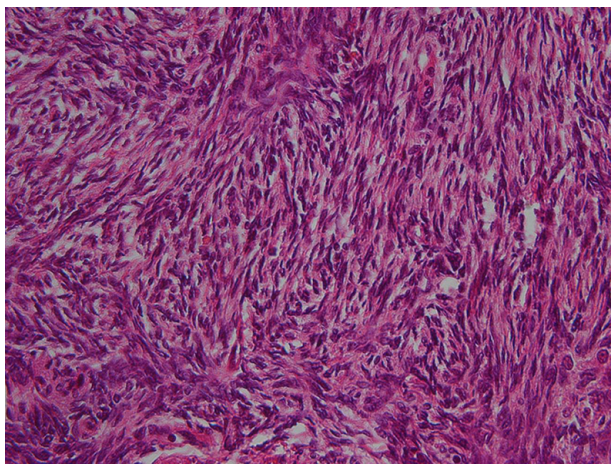


Figure 2. Areas of Viable Tumor Show Spindle Cells, Marked Pleomorphism and Frequent Mitotic Forms

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