

Leiomyosarcoma of cervical stump following subtotal hysterectomy: a case report and review of literature

Zhiqiang Liu¹, Bin Sun², Min Feng¹, Yufang Liu¹

¹Department of Obstetrics and Gynecology, Affiliated Hospital of Binzhou Medical College, Binzhou

²Emergency Department, Affiliated Hospital of Binzhou Medical College, Binzhou (China)

Summary

Background: Leiomyosarcoma that arises in the uterine cervix stump after subtotal hysterectomy is exceedingly rare. Only one case has been documented. The authors report an unusual case of leiomyosarcoma that arose in the cervix stump. **Case presentation:** A 46-year-old female presented with a one-month history of vaginal bleeding. Vaginal and ultrasonography examination revealed a mass in the cervix. The patient underwent radical resection of the cervix, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy. Chemotherapy and radiotherapy was given after the operation. The patient suffered from severe menopausal symptoms and received hormone replacement therapy. She eventually committed suicide. **Conclusion:** Given the rarity of cases of leiomyosarcoma of cervical stump, its proper treatment remains to be determined. In dealing with benign uterus diseases, we should pay adequate attention in retaining the cervix. If the cervix is retained, patients require appropriate follow-up. The authors hope that this work will benefit the fields of gynecology and oncology.

Key words: Leiomyosarcoma; Cervical stump; Subtotal hysterectomy.

Introduction

Leiomyosarcoma is a rare group of malignant tumors of mesenchymal origin and accounts for approximately 10% to 20% of all soft tissue sarcoma [1]. Leiomyosarcoma that arises in the uterine cervix stump after subtotal hysterectomy is exceedingly rare. The present authors reviewed the PubMed database for the term 'leiomyosarcoma and cervical stump', and found only one case report published in 1959 [2]. In this article, they report a case of leiomyosarcoma that arose in the cervix stump after subtotal hysterectomy in a 46-year-old female. The patient eventually committed suicide.

Case Report

A 46-year-old female with a one-month history of vaginal bleeding was admitted to the hospital on January 9, 2013. The patient had been well until a month earlier. The patient had no associated lower abdominal pain. The patient underwent subtotal hysterectomy because of uterine leiomyoma three years ago at a local hospital. The specific number of leiomyomas and their location in the uterus were not clearly recorded. The patient did not undergo further follow up in the hospital or elsewhere after subtotal hysterectomy. The patient had never undergone a cervical screening test either. Upon examination, the general conditions of the patient were good and her vitals were normal. A systemic examination revealed no abnormality. During vaginal examination, the patient's vaginal wall was smooth. A pinkish colored mass that measured approximately 5 × 4 × 3 cm was

smooth and bled when touched. The mass extended from the cervical canal. No mass could be felt in the patient's pelvis. Abdominal ultrasonography examination revealed an approximately 5 × 4 × 3 cm hypoechoic mass in the cervix, suggesting the presence of a tumor. Observations of the ovaries were inconclusive. Pancreatic, spleen, and urinary tract ultrasonography examination showed no abnormalities. The findings of chest radiography and breast sonography were normal. The admission diagnosis was as follows: 1) cervix stump tumor property to be determined; and 2) subtotal hysterectomy (three years ago). Cervical biopsy was implemented after admission and pathology examination revealed cervical leiomyosarcoma. On January 17, 2013, the patient underwent radical resection of the cervix, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy under inhalation anesthesia. During the surgery, a solid mass approximately 6.0 × 6.0 × 5.0 cm in size located between the bladder and rectum that was slightly skewed to the right was identified. A bilateral annex did not find a significant abnormality. Palpably enlarged lymph nodes were found in the right external iliac, left common iliac, bilateral inguinal, and right obturator lymph node group, whereas the other pelvic lymph nodes seemed normal. The operation was uneventful. All tissues removed were subjected to pathological examination, and the result was cervical stump leiomyosarcoma. The pathological picture showed obvious nuclear atypia and large areas of necrosis (Figures 1A and B). Immunohistochemical results showed that vimentin, SMA, and desmin were positive (Figures 1C, D, and E); CK was weakly positive (Figure 1F), whereas Ki-67 was negative. The entire layer of the cervix infiltrated the tumor. However, the tumor was not found in the surgical margin, annexes, and pelvic lymph nodes (0/23). The patient recovered well after the operation and was discharged after the abdominal

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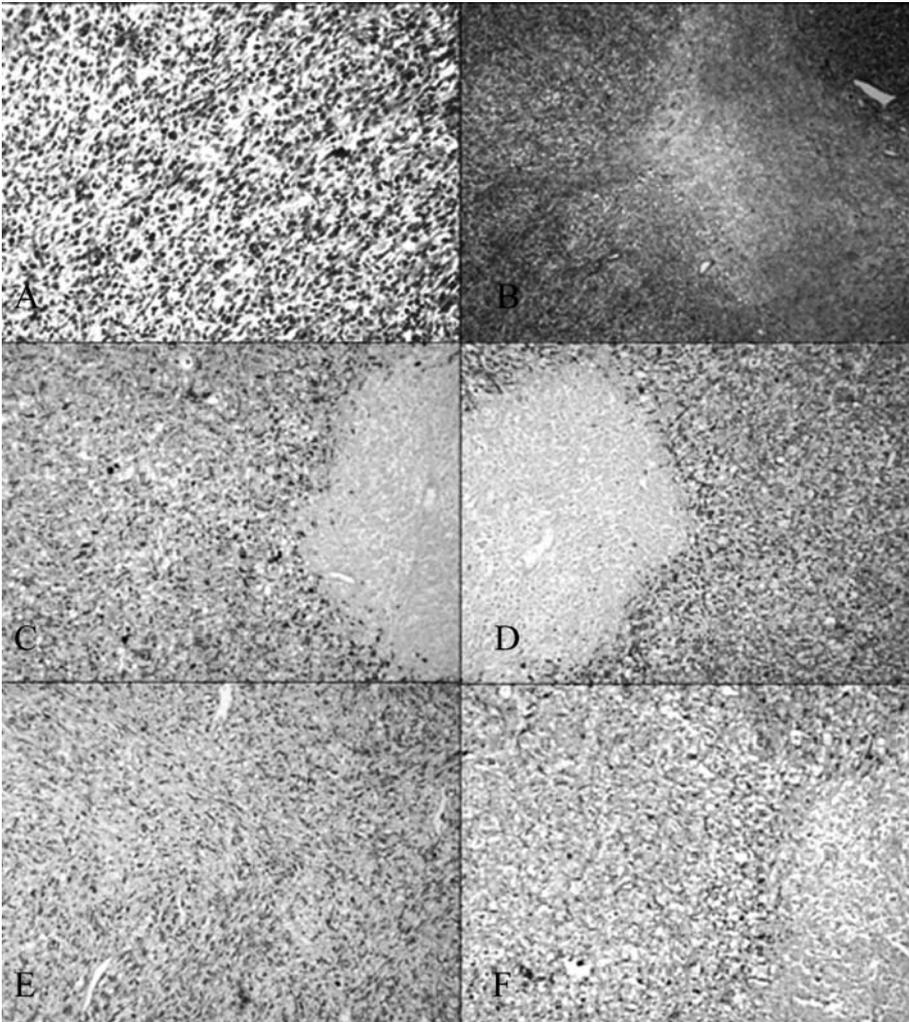


Figure 1. — A, Obvious nuclear atypia. B, Large areas of necrosis. C, Vimentin expression was positive in tumor cells. D, SMA expression was positive in tumor cells. E, Desmin expression was positive in tumor cells. F, CK expression was weakly positive in tumor cells.

incision was stitched on January 26, 2012. Adjuvant treatment in the form of chemotherapy and radiotherapy was given after the operation. Chemotherapy was implemented at the oncology department of the present hospital. She was injected with two grams of ifosfamide (D1-5) and 40 mg of cisplatin (D1-3) each cycle. The first cycle was from 16/02/13 to 20/02/13. The second cycle was from 08/03/13 to 12/03/13.

The patient developed III degree myelosuppression and II degree gastrointestinal reactions. The adverse reactions were alleviated after symptomatic treatment.

The patient received pelvic vault radiation from 23/03/13 to 08/04/13. Neutropenia occurred, so the radiation was suspended. The patient suffered from severe menopausal symptoms after gynecological surgery and received hormone replacement therapy. Sex hormone levels in her blood were examined on April 08, 2013. Results showed 58.11 IU/L follicle-stimulating hormone, 23.91 IU/L luteinizing hormone, 38.12 pmol/L estradiol, 2.19 nmol/L progesterone, 0.83 nmol/L testosterone, 105.30 mIU/L prolactin, and 448.70 nmol/L cortisol. The results indicated the presence of menopausal symptoms. The mental state of the patient had been very poor due to menopausal symptoms and from learning that she was suffering from malignant tumor. She finally committed suicide by jumping off a building on April 21, 2013.

Discussion

A brief review of cervical sarcomas: clinicopathological features, treatment, and outcome

Sarcomas of the female genital organs are rare. They account for only about 2%–3% of all malignant tumors of the genital tract [3]. Sarcomas comprise three main types: leiomyosarcoma, endometrial stromal sarcoma, and carcinosarcoma. Leiomyosarcoma is the most common type of sarcoma. Cervical sarcomas are rare neoplasms that account for only 0.005% of all cervical tumors. Most patients exhibit vaginal bleeding and palpable cervical mass at the time of diagnosis [4, 5]. Pathologically, leiomyosarcoma can be distinguished from leiomyoma based on the following features: fewer stromal fibers, larger tumor cells, increased mitotic activity, and nuclear pleomorphism [6]. A previous study [7] indicated that higher tumor grade, advanced stage of disease, and lack of primary surgical treatment are significantly associated with worse survival. Another study [8] showed that positive surgical margin, large tumor size, and advanced stage (II–IV) are associated

with the development of distant metastases and inferior survival. Given that cervical sarcomas are exceedingly rare, the optimal management of these tumors is uncertain. However, combined modality treatment that includes surgery, radiotherapy, and chemotherapy can result in prolonged survival and cure [5]. Resection of the tumor is often combined with removal of the ovaries, which may lead to serious perimenopausal symptoms for female patients. However, oophorectomy does not have an independent effect on survival [7]. Many case-control investigations have also suggested that ovarian preservation does not adversely affect survival, and that adjuvant pelvic radiation therapy does not significantly improve survival [9]. However, a different view was posited in a recent study [8]. The study showed that postoperative pelvic radiotherapy could reduce local recurrence and improve survival of patients with uterine leiomyosarcoma.

Better treatment for benign uterine diseases: total or subtotal hysterectomy

Hysterectomy is a common gynecological procedure for dealing with benign uterus diseases. Subtotal hysterectomy, which is also referred to as supracervical hysterectomy, is a common operation in which the uterus is removed and the cervix is retained. The advantages and drawbacks of total and sub-total hysterectomy remain controversial. Thakar *et al.* [10] reported that subtotal abdominal hysterectomy resulted in more rapid recovery and fewer short-term complications, but caused cyclical bleeding or cervical prolapse. After subtotal hysterectomy, 7% of women had cyclical bleeding and 2% had cervical prolapse. In another research, the author reported that compared with the different types of total hysterectomy, subtotal hysterectomy was associated with more long-term postoperative complications; by contrast, total hysterectomy was associated with more short-term complications [11]. A meta-analysis [12] showed that more women suffered from urinary incontinence and prolapse and cervical stump problems after subtotal hysterectomy than after total hysterectomy. However, subtotal hysterectomy was faster to perform, caused less preoperative bleeding, and had less intraoperative and postoperative complications. Persistent vaginal bleeding and the need for continued cervical screening appear to be the main disadvantages of subtotal hysterectomy [13]. Unfortunately, stump cancer cases showed a worse stage profile compared with cancer cases in intact uterus [13]. Therefore, the total effect of stump cancers following subtotal hysterectomy should not be neglected.

What can we learn from the case?

Patients may suffer from cervical stump problems after subtotal hysterectomy. Therefore, they need continued cervical screening. A distinguishing feature of the present case is that the patient did not undergo follow-up in a hospital or elsewhere after subtotal hysterectomy. Patients suffer from severe menopausal symptoms after oophorectomy,

and the patient in this case received hormone replacement therapy. However, sex hormone levels in the blood of the patient indicated the presence of menopausal symptoms. Unfortunately, the present patient died by suicide. She may have benefited from a more suitable dose of the drug, more care from family members, and better psychological treatment from a professional staff. Better treatment attempts can be undertaken in the future.

Conclusion

In this study, the authors presented a case of cervical stump leiomyosarcoma after subtotal hysterectomy in a 46-year-old female. She underwent treatment, including surgery, chemotherapy, and radiotherapy. Unfortunately, the patient committed suicide. The authors reviewed the relevant literature and analyzed both the clinical and pathological features of cervical stump leiomyosarcoma. They hope this paper will benefit the fields of obstetrics, gynecology, and oncology in terms of treating similar patients. We can elicit lessons from this case as follows. In view of the various medical conditions of cervical stump, we should pay adequate attention in retaining the cervix and not in dealing with benign uterus diseases. If the cervix is retained, patients require appropriate follow-up. The maximum interval for follow-ups must be six months. Follow-ups should include the following procedures: vaginal peep, pelvic examination, ultrasound examination, cervical cytology, vaginoscopy, and HPV testing. In addition, the most distressing aspect of this case is that the patient eventually committed suicide because of severe perimenopausal symptoms and her fears involving her malignant tumor. The course of treatment for this patient revealed that more effective hormone replacement therapy and more humane psychological counseling might help treat patients with malignant tumors.

In conclusion, an unusual case of a patient with subtotal hysterectomy cervical stump leiomyosarcoma was reported in this paper. The author hopes that this work will benefit the fields of gynecology and oncology.

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Address reprint requests to:

YUFANG LIU, M.D.

Department of Obstetrics and Gynecology
Affiliated Hospital of Binzhou Medical College

No.661 Huanghe Er Road

Binzhou, Shandong 256603 (China)

e-mail: lyfasb@163.com