

# Cervical leiomyosarcoma treated by radical laparoscopic approach: a case report

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## Summary

Cervical sarcomas are extremely rare neoplasms associated with younger age, advanced stage, and poorer prognosis relative to cervical carcinomas. Carcinosarcoma is the most frequently observed cervical sarcoma subtype, while leiomyosarcomas are exceedingly rare subtypes, with only a small number of cases reported in the English language literature. Cervical leiomyosarcoma patients present most commonly with abnormal vaginal bleeding, discharge, and/or abdominal and pelvic pain. Due to their rarity, data regarding these tumors typically come from case reports and small case series, and treatment strategies are often adapted from data for uterine sarcomas. Here the authors report the clinical findings from a case of unusual asymptomatic leiomyosarcoma arising in the uterine cervix and treated with a laparoscopic approach.

**Key words:** Cervical neoplasm; Laparoscopy; Leiomyosarcoma; Radical hysterectomy; Sentinel node.

## Introduction

The most common histological subtypes of cervical neoplasm is squamous cell, adenosquamous, and small-cell carcinomas. The mesenchymal tumors of uterine cervix are distinctly uncommon and heterogenous disease.

Cervical sarcomas are extremely rare neoplasms that constitute less than 1% of all cervical malignancies and are associated with younger age, advanced stage, and poorer prognosis relative to cervical carcinomas [1, 2]. Carcinosarcoma is the most frequently observed cervical sarcoma subtype, while leiomyosarcomas are exceedingly rare subtypes with only a small number of cases reported in the English language literature. Cervical leiomyosarcoma patients present most commonly with abnormal vaginal bleeding, discharge, and/or abdominal and pelvic pain.

## Case Report

A 41-year-old pre-menopausal woman (gravida 5, para 3) was admitted to this gynecological oncology department for further treatment with a confirmed diagnosis after undergoing the simple excision of a cervical tumor in another hospital. The patient had no symptoms, and the original finding occurred accidentally during the annual examination. She had amenorrhoea due to the use of the long-acting hormonal contraceptive medroxyprogesterone acetate. A spherical tumor 3 cm in diameter arising from and deforming the upper cervical lip was revealed on colposcopy. The tumor was classified as a cervical leiomyoma and simple excision of the tumor was performed.

Histopathology revealed a leiomyosarcoma with a diameter of

33×21×26 mm. The tumor consisted of atypical spindle-shaped cell fascicles with abundant eosinophilic cytoplasm (Figure 1), and fusiform nuclei with rounded ends that were hyperchromatic with coarse chromatin and prominent nucleoli. The tumor cells reacted with alpha-smooth muscle actin and h-caldesmon antibodies, as it is typical for smooth muscle tumours. The authors observed no signs of extrauterine extent of the neoplasm or angio-invasion, and there were prominent foci of coagulative cell necrosis with preserved reticulum fibres [3], and without acute inflammation. Hyaline necrosis with an intervening zone of collagen and granulation tissue between non-viable and viable tumour was also present. The mitotic index exceeded 12 figures per

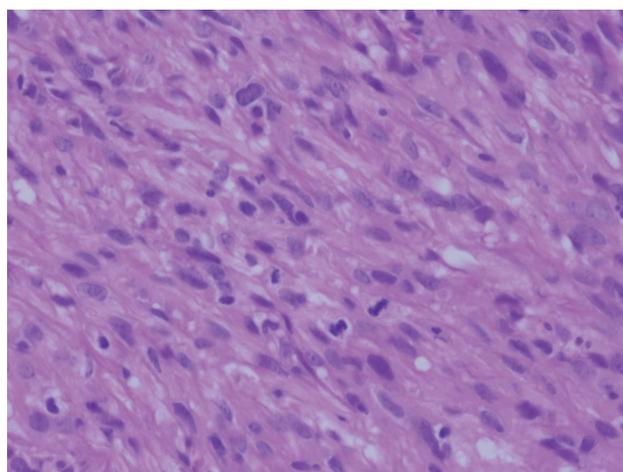


Figure 1. — Typical histological pattern of leiomyosarcoma. Tumour is hypercellular, and the cells show marked cytologic atypia and mitotic activity (Hematoxylin and Eosin, original magnification ×100).

10 high power fields and the proliferation index was approximately 20%. P16 expression was very prominent (90%) and confirmed the malignancy of the tumor, as this biomarker is expressed more strongly in leiomyosarcomas than in both usual and atypical leiomyoma [4].

Gynecological examination performed in the authors' department following the excision, did not reveal any residual tumor, or indication of vaginal or parametrial involvement. The clinical stage was determined to be FIGO I. The sonography expert reported an absence of residual cervical tumor, parametrial involvement, and suspicious lymph nodes in the pelvic region.

Following the examination, exclusion of distant metastases, and after detailed counseling with respect to the age and desires of the patient, the authors recommended total radical laparoscopic hysterectomy at the extent of the B1 radicality according to Querleu-Morrow classification, with bilateral salpingo-oophorectomy and pelvic lymphadenectomy with sentinel node detection. The surgery and postoperative period were uneventful. Final histology report indicated no residual tumor in the cervix, no parametrial involvement, and no metastatic involvement of either sentinel or non-sentinel lymph nodes. The patient remains disease-free more than four years after the treatment.

## Discussion

Primary sarcomas arising in the cervix are extremely rare. According to Wright *et al.*, of 1,583 newly diagnosed invasive cervical tumors, only eight were identified as cervical sarcomas [1]. Cervical sarcomas therefore accounted for only 0.005% of cervical malignancies, and of those eight cervical sarcomas, only one was leiomyosarcoma, while the remaining seven were carcinosarcomas. Bansal *et al.* reported a total of 33,074 women with identified invasive cervical neoplasm, including 323 (1%) patients with cervical sarcoma. Carcinosarcoma was the most common type among women with cervical sarcomas, and accounted for 40% of cases while adenocarcinomas and leiomyosarcomas each accounted for 21% [2]. These large studies of cervical sarcomas also included carcinosarcomas, as they are considered to be types of metaplastic carcinomas rather than sarcomas.

Patients with cervical sarcomas tend to be younger, have larger tumors, and have a more advanced stage disease relative to squamous cell carcinoma and adenocarcinoma patients [2]. In a review of 208 patients with leiomyosarcoma of the uterus, Guintoli *et al.* found that stage, older age (> 51 years), postmenopausal status, and larger tumor size (> 5 cm) were associated with more frequent and significantly reduced survival likelihood [5].

In a retrospective analysis, Wright and Khosla determined that all patients with cervical sarcoma presented with vaginal bleeding [1, 6]. In the present case, the patient was asymptomatic and the cervical tumor was detected accidentally during an annual gynecological examination.

Due to the infrequent occurrence of these neoplasms and inconsistent treatment modalities in case reports and small case series studies, recommendations regarding the treatment of cervical leiomyosarcoma have not been proven.

Total abdominal hysterectomy with bilateral salpingo-oophorectomy is performed in most cases of leiomyosarcoma restricted to the cervix [1, 6-8].

In the Gynecologic Oncology Group Study (GOG) study reported by Major *et al.*, lymph node metastases were identified in only 3.5% of patients with clinically localized uterine leiomyosarcomas at the time of surgical staging [9], and after 30 years, the need for lymph node dissection in patients with leiomyosarcoma remains unsubstantiated. In Bansal *et al.*'s report of 26 patients with leiomyosarcoma who underwent lymphadenectomy, none showed nodal metastases [2]. Lymphatic mapping and sentinel lymph node detection could provide important information about the status of pelvic lymph nodes, including detection of micrometastases and isolated tumor cells in these cases, allowing for the consideration of limited surgical radicality [10].

Radical surgery with a laparoscopic approach was chosen in the present case due to the cervical localization of the tumor and possible lymphatic spread through the parametria to the pelvic lymph nodes. The authors performed a total laparoscopic radical hysterectomy of B1 type radicality according to Querleu-Morrow classification, with bilateral salpingo-oophorectomy and pelvic lymphadenectomy with sentinel node detection. The postoperative course was uneventful and the patient was discharged on the fifth postoperative day. The final histology report revealed no residual tumor in the cervix, no parametrial involvement, and no metastatic involvement of sentinel and non-sentinel lymph nodes. No adjuvant therapy was indicated due to these promising histopathological factors. The patient remains disease-free 52 months after the surgical treatment.

There is no clear evidence that adjuvant radiation therapy or chemotherapy would bring benefit for uterine sarcoma patients. However, most patients with advanced disease are considered to be chemotherapy candidates due to the known tendency towards hematogenous metastasis. To the present authors' knowledge, this is the first case of cervical leiomyosarcoma treated by the laparoscopic approach in the literature.

In conclusion, total laparoscopic radical hysterectomy with sentinel lymph node detection is a feasible and comprehensive treatment for patients with low-stage cervical leiomyosarcoma, and the patient benefits from this minimally invasive approach.

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