



# Early-stage cervical leiomyosarcoma and carcinosarcoma with long-term disease-free survival after surgery alone: a dual case report

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## ABSTRACT

Cervical leiomyosarcoma and cervical carcinosarcoma are extremely rare malignancies, and optimal management strategies remain undefined. In early-stage disease, the benefit of adjuvant therapy is controversial, raising concerns about potential overtreatment. We report two patients with early-stage primary cervical sarcomas who presented with similar clinical findings but were diagnosed with different histopathological subtypes. A 47-year-old woman presented with abnormal vaginal bleeding and a polypoid cervical mass and was diagnosed with cervical leiomyosarcoma following complete excision. A 66-year-old postmenopausal woman presented with postmenopausal bleeding and was diagnosed with cervical carcinosarcoma after comprehensive surgical staging. In both cases, preoperative imaging demonstrated disease confined to the cervix, and final pathology confirmed FIGO stage IB2 tumors with negative surgical margins and no lymph node or distant metastases. Multidisciplinary tumor board evaluation was performed, and adjuvant chemotherapy or radiotherapy was not administered in either patient. Despite the aggressive histological features traditionally associated with these tumor types, both patients achieved sustained long-term disease-free survival at 2 and 4 years of follow-up, respectively. These cases suggest that selected patients with early-stage cervical sarcomas may achieve favorable outcomes with surgery alone and support a risk-adapted, individualized approach to adjuvant treatment decisions to avoid potential overtreatment.

**Keywords:** Cervical sarcoma, leiomyosarcoma, carcinosarcoma, surgery alone, overtreatment, risk-adapted treatment, case report

## INTRODUCTION

Sarcomas of the uterine cervix are extremely rare, accounting for less than 1% of all cervical malignancies.<sup>1</sup> Among these, cervical leiomyosarcoma (CLMS) and cervical carcinosarcoma (CCS) represent particularly uncommon histological subtypes and pose significant diagnostic and therapeutic challenges. Due to their rarity, available evidence is limited to case reports and small case series, and no standardized management guidelines exist.

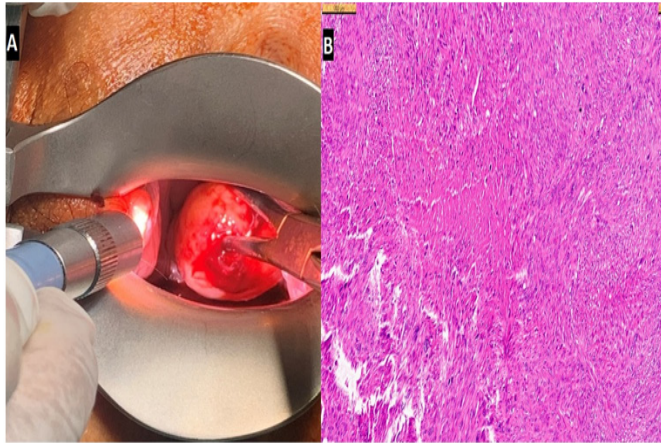
Leiomyosarcoma commonly arises in soft tissue and visceral organs; however, primary involvement of the uterine cervix is extremely uncommon. A 2020 literature review identified only 40 reported cases of primary CLMS worldwide.<sup>2</sup> Patients typically present with vaginal bleeding and a cervical mass, and the disease predominantly affects peri- and postmenopausal women.<sup>3</sup> Carcinosarcoma is a biphasic malignancy composed of both epithelial and mesenchymal components and most frequently arises in the uterine corpus.<sup>4</sup> Primary CCS is exceedingly rare, accounting for

approximately 0.005% of cervical cancers, with only 70 cases reported in the English literature by 2013.<sup>5</sup> Similar to CLMS, the most common clinical findings are vaginal bleeding and a protruding cervical mass, and most patients are postmenopausal.<sup>6</sup> There is no established consensus regarding the optimal management of CLMS, including the roles of lymphadenectomy and adjuvant therapy. Likewise, treatment strategies for CCS remain unclear and are largely extrapolated from data on uterine carcinosarcoma.<sup>6</sup>

Despite distinct histopathological features, CLMS and CCS may present with similar clinical findings, making accurate pathological diagnosis essential for appropriate management. In early-stage disease, the role of adjuvant therapy remains controversial, and concerns regarding potential overtreatment have emerged. In this report, we present two cases of early-stage cervical sarcoma with similar clinical presentations but different histological diagnoses, both achieving favorable long-term outcomes following surgical management alone.

## CASE 1

A 47-year-old woman (G4P2) presented with persistent vaginal bleeding for two months. Her medical and family histories were unremarkable. Cervical smear tests performed during the previous year were normal. On gynecological examination, a hemorrhagic polypoid mass approximately 3 cm in diameter was observed protruding from the cervix (**Figure 1**).



**Figure 1.** Tumor appearance of the case 1. (A) Macroscopic view of the tumor (B) Tumor necrosis and mitotic figures in smooth muscle bundles with atypical, spindle-shaped morphology

A cervical punch biopsy was attempted before surgery; however, the specimen was nondiagnostic due to extensive necrosis and hemorrhage. Therefore, complete surgical excision of the cervical mass was performed together with endometrial curettage. The excised mass measured 4×3×3 cm. Histopathological examination revealed moderate cellular atypia, tumor necrosis and a mitotic activity of 9 per 10 high-power fields, consistent with a diagnosis of leiomyosarcoma. Immunohistochemical staining demonstrated positivity for smooth muscle markers including SMA and desmin and negativity for cytokeratin markers, supporting the diagnosis of leiomyosarcoma. No additional molecular or genetic analyses were performed in these cases, which represents a limitation of the present report. No malignant cells were detected in the endometrial curettage specimen. Preoperative imaging with contrast-enhanced computed tomography of the chest, abdomen and pelvis showed no evidence of metastatic disease. Pelvic magnetic resonance imaging demonstrated a lesion confined to the cervix, with no evidence of parametrial, uterine or extra-cervical extension.

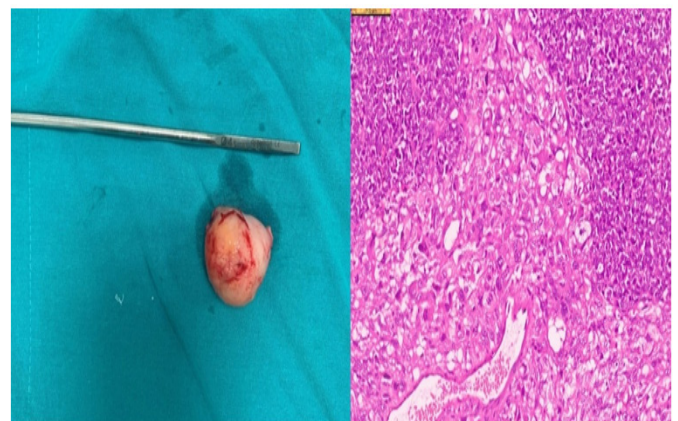
The patient subsequently underwent comprehensive surgical staging, including total hysterectomy, bilateral salpingo-oophorectomy, omentectomy and pelvic and para-aortic lymph node dissection. Although routine lymphadenectomy is not universally recommended in leiomyosarcoma, comprehensive staging including pelvic and para-aortic lymph node dissection was performed in this case due to the initial diagnostic uncertainty and to exclude occult metastasis as part of institutional staging practice. Final pathological examination showed no residual tumor in the surgical specimen. All surgical margins were negative and no lymph node or distant metastases were identified. The final stage was determined as FIGO stage IB2.

The case was discussed in a multidisciplinary tumor board including gynecologic oncology, pathology and medical oncology specialists. Given that the tumor was confined to the cervical polyp, completely excised, with negative margins and no evidence of residual or metastatic disease, adjuvant chemotherapy or radiotherapy was not recommended. The patient has remained disease-free for two years and is followed regularly every three months with pelvic examination and imaging.

## CASE 2

A 66-year-old postmenopausal woman (G2P2) presented with a two-month history of postmenopausal spotting. Her medical and family histories were unremarkable. Previous cervical cytology and HPV test results were not available in the electronic medical records. On gynecological examination, a necrotic and bleeding mass approximately 3 cm in diameter was observed arising from the cervical canal. Bimanual examination revealed no parametrial involvement. Transvaginal ultrasonography demonstrated that the lesion was confined to the cervix and was independent of the uterus and adnexal structures.

A cervical punch biopsy performed prior to surgery revealed atypical epithelial proliferation with spindle cell components, raising suspicion for a mixed malignant tumor. Preoperative imaging with contrast-enhanced computed tomography of the chest, abdomen, and pelvis showed no evidence of distant disease. Pelvic magnetic resonance imaging demonstrated a cervical lesion without extension to the parametrium, uterus, or adjacent pelvic structures. The patient subsequently underwent comprehensive surgical treatment, including radical hysterectomy, bilateral salpingo-oophorectomy, omentectomy and pelvic and para-aortic lymph node dissection. The excised tumor measured 3.5×2×2 cm. Final histopathological examination confirmed the diagnosis of CCS. The tumor was confined to the cervix, did not extend to the vagina, and showed no lymphovascular invasion. Surgical margins were negative. The omentum was benign, and no lymph node metastases were identified. The final stage was determined as FIGO stage IB2. Immunohistochemical analysis demonstrated strong positivity for p16, pan-cytokeratin, CAM5.2, and PAX8, with desmin positivity in the sarcomatoid component. p53 was positive in both epithelial and mesenchymal components, while p40 was negative (**Figure 2**).



**Figure 2.** Tumor appearance of the case 2. (A) Macroscopic view of the tumor (B) Showing a high-grade malignant tumor with biphasic morphology (H&E, 40x)

The case was discussed at a multidisciplinary tumor board. Given the aggressive biological behavior of carcinosarcoma, adjuvant treatment with four cycles of carboplatin–paclitaxel chemotherapy and external beam radiotherapy was recommended. However, the patient declined further treatment. She has remained disease-free for four years and is followed regularly with pelvic examination and imaging every three months for the first two years and every six months thereafter.

## DISCUSSION

Primary sarcomas of the uterine cervix, including CLMS and CCS, are exceedingly rare and account for less than 1% of all cervical malignancies. Because of this rarity, available evidence is largely derived from case reports and small case series, which leads to heterogeneous treatment approaches and a lack of consensus guidelines. In the present report, we describe two early-stage cervical sarcomas with distinct histopathological subtypes: one CLMS and one CCS. Both patients were treated with comprehensive surgical staging alone without adjuvant therapy and remain disease-free at two and four years of follow-up. These outcomes provide meaningful clinical evidence regarding the ongoing debate on the necessity of adjuvant treatment in selected early-stage cases.

Surgery remains the cornerstone of treatment for both CLMS and CCS. In the largest systematic review and meta-analysis of CLMS, Kılıç et al.<sup>2</sup> demonstrated that hysterectomy was an independent prognostic factor for disease-specific survival, whereas the extent of surgery did not significantly influence recurrence. Similarly, Bansal et al.<sup>7</sup> reported no lymph node metastases among patients who underwent lymphadenectomy, and Kılıç et al.<sup>2</sup> observed no nodal involvement in most patients who had lymph node dissection. These findings suggest that routine lymphadenectomy may not be necessary in CLMS and should be reserved for patients with bulky or radiologically suspicious lymph nodes. In localized CCS, surgical management typically includes radical hysterectomy with bilateral salpingo-oophorectomy and pelvic lymph node dissection. Radical surgery is strongly recommended for stage IB disease, which accounts for nearly half of reported cases, whereas cytoreductive surgery is generally reserved for advanced disease or for patients who are not candidates for radical surgery.<sup>8</sup>

To date, no studies have defined an optimal adjuvant treatment strategy for cervical sarcomas. In CLMS, reported adjuvant approaches include chemotherapy alone or combined chemoradiotherapy, yet available data have failed to demonstrate a clear survival benefit. Given the substantial toxicity associated with systemic therapy, the role of adjuvant treatment in early-stage CLMS remains uncertain.<sup>9</sup> In CCS, some authors advocate aggressive adjuvant treatment. A meta-analysis by Comert et al.<sup>10</sup> recommended adjuvant radiotherapy with or without chemotherapy for locally advanced disease and reported improved disease-free survival in early-stage cases treated with multimodal therapy. However, these recommendations are largely extrapolated from uterine carcinosarcoma data. In the review by Shu et

al.,<sup>6</sup> most reported patients received radiotherapy, but the absence of randomized or well-designed comparative studies limits any definitive conclusions regarding its impact on survival. Similarly, although platinum-based chemotherapy regimens are frequently used, their true efficacy remains difficult to assess.

Despite the frequent use of adjuvant therapy in published reports, favorable outcomes have also been observed in selected early-stage patients treated with surgery alone. In CLMS, Doshi et al.<sup>11</sup> reported recurrence-free survival in a patient managed without adjuvant therapy, whereas Khosla et al.<sup>12</sup> described early mortality in an older untreated patient, supporting evidence that age may be an independent prognostic factor. In CCS, Sharma et al.<sup>13</sup> reported long-term disease-free survival in two patients with stage IB disease managed without adjuvant therapy, and Lin et al.<sup>14</sup> described a similar outcome in a patient with stage IB1 disease treated with surgery alone. These observations suggest that disease stage and patient-related factors may be more relevant determinants of outcome than routine adjuvant therapy and that surveillance after surgery may be appropriate in carefully selected early-stage cases.

## CONCLUSION

Primary sarcomas of the uterine cervix, including CLMS and CCS, are exceedingly rare malignancies with overlapping clinical presentations. Due to limited evidence derived mainly from case reports, standardized treatment algorithms are lacking. While surgery remains the cornerstone of management in early-stage disease, the role of adjuvant therapy is undefined. These observations may contribute to the limited existing literature suggesting that surgery alone could be considered in carefully selected early-stage cases; however, definitive recommendations require larger collaborative studies. Multicenter collaborative studies are needed to refine evidence-based management strategies.

## ETHICAL DECLARATIONS

### Informed Consent

Written informed consent was obtained from the patient(s) included in this report. Signed consent forms are retained by the authors and are available upon request.

### Peer Review Process

This report underwent external peer review.

### Conflict of Interest

The authors declare no conflicts of interest.

### Financial Disclosure

This case report did not receive any financial support.

### Author Contributions

Concept: B.M.S.; Design: B.M.S., H.K.Ç., E.A.; Materials: B.M.S., H.K.Ç., E.A.; Data Collection and/or Processing: B.M.S., H.K.Ç., E.A.; Writing the Article: B.M.S.; Critical Review: B.M.S., H.K.Ç., E.A.; Final Approval: B.M.S., H.K.Ç., E.A.

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