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Case Report

High-grade cervical leiomyosarcoma: a case report

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ABSTRACT

Cervical leiomyosarcomas represent less than 1% of all malignant cervical neoplasms, predominantly affecting perimenopausal women, with an average age of diagnosis at 46 years. This article presents a case involving a 45-year-old female patient admitted to the gynecology department due to pyelonephritis. During initial evaluation, a cervical tumor was incidentally discovered. Histopathological analysis revealed findings consistent with high-grade leiomyosarcoma, not otherwise specified (NOS). The patient underwent surgical intervention, which included sigmoid resection, type III Piver hysterectomy, mechanical side-to-side colorectal anastomosis, appendectomy, placement of JJ stents, and terminal ureterovesical anastomosis. Leiomyosarcomas are associated with a poor prognosis, even when diagnosed at an early stage. The most prevalent symptom is abnormal vaginal bleeding. Surgical intervention is the primary treatment modality for this condition, with the objective of achieving an R0 resection, which entails the complete removal of the tumor.

Keywords: Sarcoma, Leiomyosarcoma, Malignant tumor, Vaginal bleeding

INTRODUCTION

Female genital tract sarcomas represent approximately 3% of all gynecological malignancies. Among these, cervical leiomyosarcomas are exceedingly rare, and constitute less than 1% of all cervical malignant tumors. These tumors predominantly affect perimenopausal women, with the average age at the time of diagnosis around 46 years.[1] Although leiomyosarcoma is one of the most common non-epithelial malignancies arising in soft tissues and somatic organs, cases arising from the cervix are extremely rare.[2]

A variant of these tumors can originate in the cervix or in the uterine body, specifically in the cervical stroma or myometrium without involving the endometrium. They are brown, red, or yellow with a tendency towards white, in addition to being friable and hemorrhagic. Cystic changes and necrosis may also be present. Regarding their histopathology, sheets of uniform cells with granular eosinophilic cytoplasm and striated, irregular nuclei are occasionally found, either centrally or eccentrically located, with vesicular chromatin and prominent nucleoli, which may be associated with edema or myxoid changes. Spindle cells are usually composed of nuclei of the same type, discrete nucleoli, and abundant wavy eosinophilic cytoplasm.[3]

CASE REPORT

A 45-year-old female patient with obesity and a history of hypothyroidism since the age of 17, treated with levothyroxine; her surgical history includes two cesarean sections. Her gynecological and obstetric history highlights a last cervical cytology in 2021, which showed no lesions or malignancy. The patient presented with

abnormal transvaginal bleeding, suprapubic pain, and dysuria, which were managed with analgesics and oral hydroxyprogesterone without adequate response. Symptoms persisted, and four weeks later, she developed hyporexia and unintended weight loss of approximately 6 kg over 3 weeks, as well as recurrent urinary tract infections, leading to multiple emergency department visits (Figure 1). On physical examination, the patient was conscious, oriented, and cooperative. Directed examination revealed a palpable lesion detected bimanually during vaginal examination, with an indurated, fixed, non-friable consistency, painful on palpation, seemingly originating from the cervix, approximately 10×15 cm in size, with involvement of the right and left parametria. Upon completion, the examining glove showed traces of bleeding.

A pelvic ultrasound was performed, revealing a uterus with altered morphology and a tumor mass in the cervix extending towards the vagina and rectum, with a tumor measuring 10×15 cm originating from the uterus. Subsequently, a simple and contrast-enhanced abdominopelvic CT scan was conducted, which reported a bladder with minimal distension and no evidence of intra- or extraluminal lesions; a uterus in its usual position, with the cervix showing increased dimensions due to an ovoid-shaped image with well-defined borders and heterogeneous density due to areas of lower density, suggestive of necrotic areas (Figure 2). After the administration of contrast medium, a heterogeneous enhancement predominantly at the periphery was identified, causing cranial displacement of the uterus and anterior displacement of the bladder, with approximate dimensions of 162×96×86 mm in the longitudinal, transverse, and anteroposterior axes, respectively (Figure 3 and 4).

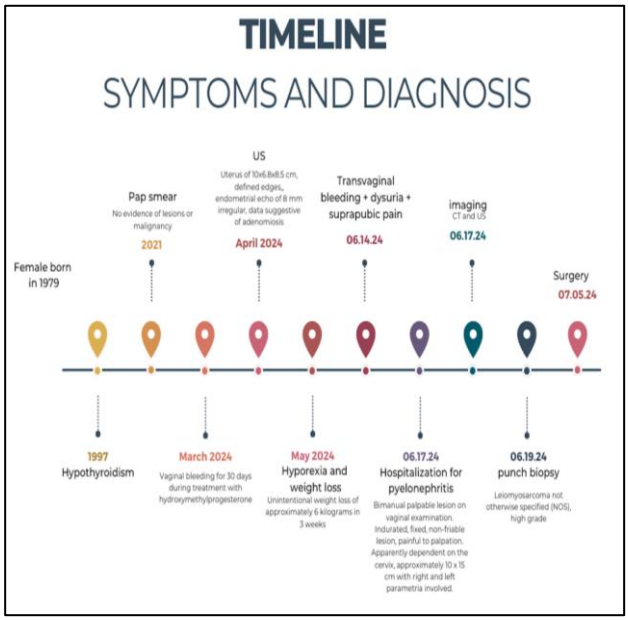

Figure 1: Disease progression: symptoms and diagnosis.

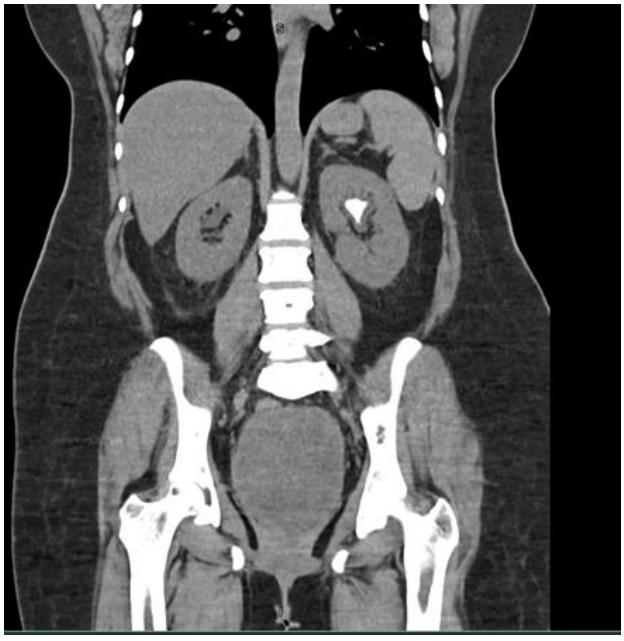

Figure 2: Non-contrast CT scan of the abdomen and pelvis in coronal view: Identified left renal pelvis dilation as well as major calyceal group dilation. In the pelvic cavity, at the level of the cervix, there is an increase secondary to an ovoid-shaped image with well-defined borders and heterogeneous density due to areas of lower density suggestive of necrosis. Approximate dimensions are 162×96×86 mm in the longitudinal, transverse, and anteroposterior axes, respectively.

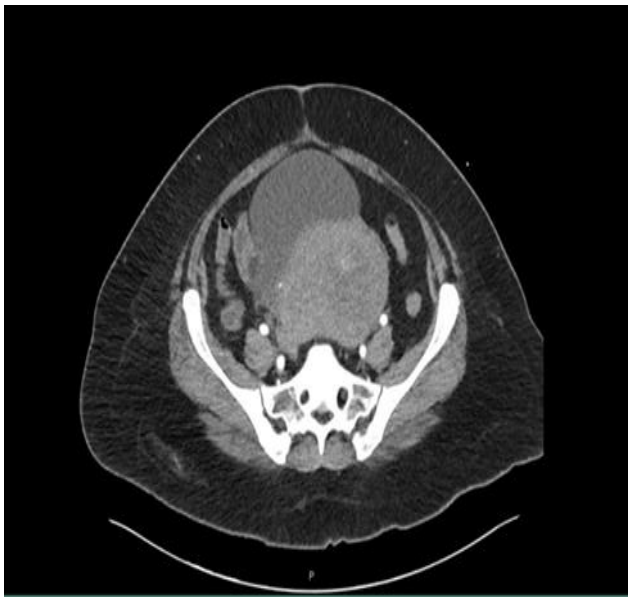

Figure 3: Contrast-enhanced axial CT scan of the pelvic cavity: Solid nodular image of heterogeneous density with well-defined lobulated borders. After contrast medium administration, there is heterogeneous enhancement, revealing a hypodense component with liquid density, which is consistent with hyaline degeneration.

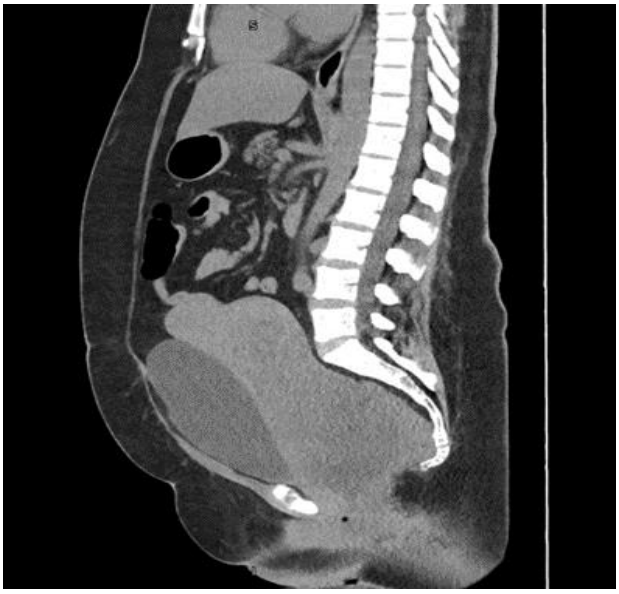

Figure 4: Contrast-enhanced sagittal CT scan: Bilobulated heterogeneous image in uterine topography with an adequate pericecal and vesical interface, displacing adjacent organs due to its size.

A punch biopsy was performed, resulting in a histopathological finding of fragmented tissue with ischemic changes, recent hemorrhage, and marked vascular congestion. In the viable areas, spindle cell proliferation was identified, growing in irregular fascicles composed of medium to large-sized cells with ovoid to round nuclei and coarse chromatin. Some cells exhibited pleomorphism. Focal tumor necrosis was observed, and mitotic activity was noted at up to 7 mitoses per 10 high-power fields. No lymphovascular invasion was identified. These morphological features were consistent with high-grade leiomyosarcoma (Figure 5).

Figure 5: Cervical tumor, with morphological data compatible with high-grade leiomyosarcoma.

The patient underwent surgical treatment, which included a conversive sigmoidectomy, a Piver type III hysterectomy, a mechanical lateral-to-lateral colorectal anastomosis, appendectomy, placement of JJ stents, and a terminal-to-lateral ureterovesical anastomosis.

Initially, a laparoscopic approach was attempted, but a tumor was found in the pelvic cavity dependent on the uterus. Due to the tumor's characteristics, the decision was made to convert the surgery to an open approach.

A median incision, approximately 20 cm in length, was made infra- and supraumbilically. A tumor, measuring approximately 25×15×12 cm, was observed, dependent on the uterus. The tumor was adherent anteriorly to the bladder, laterally to the iliac vessels, to both ureters, and posteriorly to the rectum.

During the hysterectomy, a vaginal examination confirmed that the adnexa were free, so oophorectomy and salpingectomy were deferred. The remaining intestinal edges from the sigmoidectomy were anastomosed using a 60 mm linear stapler, and a pneumatic test was performed to confirm the integrity of the anastomosis. The remaining edges of the ureters were repaired and invaginated into the bladder, and double-J stents were subsequently placed. An appendectomy was performed using Enseal and then sutured with 2-0 silk. Six Gelfoam sponges were placed in the pelvic cavity (rectum, vaginal dome), and an additional six were placed in the bed of the iliac arteries. Two Penrose drains (½ inch) were inserted, one into the pelvic cavity and the other into the Retzius space, both externalized to the right flank. During the procedure, a blood loss of 1200 cc occurred, and the patient received 3 units of packed red blood cells and 2 units of fresh frozen plasma.

DISCUSSION

Symptoms typically include abdominal distension, palpable cervical or abdominal mass, pelvic pain, vaginal protrusion, obstructive symptoms such as increased urinary frequency and urinary retention, weight loss, and abnormal vaginal bleeding, with the latter being the most common symptom.[1,2,4,9] In this case, the patient exhibited most of the previously described symptoms, complicated by pyelonephritis secondary to urinary retention.

Imaging tests play a crucial role in diagnosis, including computed tomography (CT) and magnetic resonance imaging (MRI), which have been reported as highly useful for differentiating between ordinary leiomyomas, their variants, and leiomyosarcomas.[5] However, definitive diagnosis is achieved through histopathological and immunohistochemical evaluation. In this regard, authors note that spindle-shaped cells with elongated and ovoid nuclei, prominent nucleoli, and abundant eosinophilic cytoplasm may be present. Additionally, mitotic activity is marked with a mean index of 10/10 high-power fields in the epithelioid component.[3] These characteristics were observed in this case. Differential diagnosis for this pathology includes squamous cell

carcinoma and other cervical sarcomas such as rhabdomyosarcoma, liposarcoma, primitive neuroectodermal tumor, and malignant peripheral nerve sheath tumor.[2]

Currently, surgical management remains the cornerstone of treatment for leiomyosarcoma. As such, total abdominal hysterectomy with bilateral salpingo-oophorectomy with wide negative margins is considered the gold standard of treatment.[5-7] Despite this, multiple surgical techniques have been described, such as extrafascial hysterectomy, modified radical hysterectomy, and radical hysterectomy with or without retroperitoneal lymph node dissection. However, the type of hysterectomy does not influence the oncological outcome as long as there are clear surgical margins.[8] We are awaiting definitive histopathology results to determine the success of the surgery.

Leiomyosarcomas have an unfavorable prognosis even at an apparently early stage.[4] Similar to other histological variants of sarcoma, the mode of spread is predominantly hematogenous, with the lungs being the most common site of metastasis.[9] According to imaging study results, the patient did not present with metastasis and was pulmonarily asymptomatic, without need for supplemental oxygen.

Significant independent predictors of local recurrence are size and margin, while predictors for distant recurrence are size and grade. Therefore, for patients at high risk of developing leiomyosarcoma, it is necessary to implement additional adjuvant or neoadjuvant therapy alongside surgical treatment.[6] We are currently awaiting the results of immunohistochemistry in addition to the final histopathology report to determine the most appropriate cytotoxic treatment for our patient.

The role of chemotherapy has been extrapolated from uterine body sarcomas; the most commonly used drugs are doxorubicin and ifosfamide. This regimen has an overall response rate of 30% in advanced and metastatic forms of the disease. Other chemotherapy drugs used include cyclophosphamide, gemcitabine, and docetaxel, which have response rates of approximately 30%. Radiotherapy is generally administered in the adjuvant setting to prevent local regional recurrence. The radiotherapy regimen typically delivered is an external beam radiotherapy dose of 50 Gy in 25 fractions over 5 weeks with 2 Gy per fraction, followed by brachytherapy consisting of 2-3 brachytherapy sessions that can also be administered during EBRT to reduce the overall treatment duration, impacting locoregional control rates.[9] Fortunately, in our institution, all the aforementioned drugs and radiotherapy are available.

Therefore, it should be considered that the treatment of this rare histopathological variant affecting the cervix involves multimodal management consisting of surgery, chemotherapy, and radiotherapy, as well as pre- and post-surgical rehabilitation and pre- and post-cytotoxic treatment rehabilitation. This approach increases survival rates and quality of life in these patients.[10,11]

CONCLUSION

Cervical leiomyosarcoma is a rare and highly aggressive entity for which no clear consensus has been established regarding optimal surgical and adjuvant treatment. We report this case to add to the existing literature due to the rarity of this pathology. With this case, we aim to emphasize that the diagnostic and therapeutic approach for patients with these pathologies should be conducted in a tertiary care center, as it is crucial to have a multidisciplinary team for proper management. Additionally, efforts should be increased in educating the population about screening studies and primary care physicians about rare pathologies to facilitate early diagnosis of this histological subtype.

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