

CASE REPORT: CLINICAL CHALLENGES IN THE MANAGEMENT OF PRIMARY AND RECURRENT BROAD LIGAMENT LEIOMYOSARCOMA—A COMPARATIVE SERIES OF TWO PATIENTS

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ABSTRACT

Background: Primary broad ligament leiomyosarcoma is an ultra-rare malignancy representing less than 1% of gynecological cancers. Due to its scarcity, standardized therapeutic guidelines do not exist.

Case Presentations: We report two distinct recurrent cases from our tertiary center (2021–2025). Case 1 involves a 28-year-old premenopausal female presenting with a giant 25 cm retroperitoneal mass causing severe hydronephrosis. During radical baseline excision, an accidental internal iliac artery laceration was successfully repaired. Following fertility preservation and chemotherapy, she developed a localized pelvic side wall recurrence three years later, successfully treated via R0 secondary cytoreduction and delayed hysterectomy. She remains in sustained complete remission as of May 2025. Case 2 describes a 64-year-old postmenopausal female whose primary tumor was extracted via fragmentation at a regional facility. Her early recurrence was radiologically misidentified as a benign postoperative lymphocele. This diagnostic delay allowed progression to extensive multi-visceral peritoneal carcinomatosis, requiring aggressive secondary multi-visceral debulking and bowel resection followed by gemcitabine-docetaxel chemotherapy.

Conclusions: Broad ligament leiomyosarcoma demands intact tumor extraction and negative margins. Clinicians must evaluate postoperative pelvic fluid collections with high suspicion to avoid misdiagnosing early recurrence. Complete secondary cytoreduction offers optimal local control.

KEYWORDS: Broad ligament leiomyosarcoma, Recurrent sarcoma, Secondary cytoreduction, Peritoneal carcinomatosis, Case series

INTRODUCTION

Broad ligament leiomyosarcoma is a rare malignant tumor arising from smooth muscle within the broad ligament, a peritoneal fold that supports the uterus [1]. As a highly aggressive form of uterine sarcoma, it often presents with nonspecific symptoms like pelvic pain, mass effect, or abnormal bleeding. Due to its rarity, diagnosis can be challenging and may require imaging and histopathological analysis [2]. Treatment typically involves surgical excision, often combined with adjuvant radiotherapy or chemotherapy. Despite advances in management, the prognosis remains guarded, underscoring the importance of early detection and comprehensive, multidisciplinary care to improve patient outcomes [3].

This case series stands out as an exceptionally rare and clinically significant contribution to gynecologic oncology literature. Primary leiomyosarcoma (LMS) of the broad ligament is an exquisite rarity, accounting for less than 1% of all gynecological malignancies, with fewer than 30 cases documented in the global English medical literature [1,4]. To diagnose a true broad ligament LMS, the malignancy must fulfill Gardner's criteria [5], meaning it must develop entirely separate from and exhibit no connection to either the uterus or the ovaries [1,4].

Case 1

A 28-year-old single, nulliparous female with no significant past medical or surgical history presented to KKUH. She reported a progressive, severely painful pelvic-abdominal mass accompanied by rapid abdominal distension. Her family history was negative for soft-tissue sarcomas, early-onset gynecological malignancies, or known hereditary cancer syndromes. The abdomen was markedly distended by a firm, minimally mobile pelvic-abdominal mass extending into the mid-abdomen. Deep palpation elicited significant tenderness across the right lower quadrant and suprapubic regions.

The Gynecological ultrasound showed a normal-sized anteverted uterus with a 9 mm endometrial thickness and a normal left ovary. A large pelvic-abdominal mass measuring 19.4 x 10 cm was identified posterior to the uterus. The pouch of Douglas was free of fluid, but the right ovary was not visible. CT scans revealed a clear thoracic cavity with no pulmonary metastasis. A giant retroperitoneal pelvic mass was displacing and compressing the right urinary tract, causing moderate-to-severe hydronephrosis and hydronephrosis (Figure 1).

The patient underwent an urgent exploratory laparotomy through an extended midline incision. Exploration revealed a large 25 cm, highly vascular retroperitoneal mass originating from the right broad ligament, with the uterus, fallopian tubes, and ovaries appearing normal. During dissection, severe tumor adherence to the right

pelvic sidewall led to an accidental laceration of the right internal iliac artery. The vascular surgery team was called in, and proximal control of the right common iliac vessels was achieved. The artery was successfully repaired with primary suturing, restoring blood flow. The procedure resulted in an estimated blood loss of 3 liters, causing temporary hemorrhagic shock. Hemodynamic stability was regained after transfusing 6 units of PRBC, 6 units of platelets, and 6 units of FFP. Hemostasis was secured with TachoSil sponges, surgical powder, and Aresta powder. A Jackson-Pratt drain was placed in the pelvic cavity. The postoperative recovery was stable; abdomen soft with mild tenderness, initial dark blood drain, normal vital signs, and confirmed right kidney recovery.

The gross examination revealed an encapsulated tumor weighing 1913 grams and measuring 22.0 x 17.0 x 10.0 cm, with a white-tan cut surface showing hemorrhage and necrosis (~5%). Microscopy showed spindle cell malignancy with severe nuclear atypia, hyperchromatism, and a high mitotic rate. Margins were negative, with a close margin of 0.1 cm; focal lymphovascular invasion was noted. Immunohistochemistry was positive for Desmin and SMA, confirming a diagnosis of high-grade primary right broad ligament leiomyosarcoma.

Given the high-grade features, close margins, and focal LVI, the Multidisciplinary Team (MDT) recommended fertility preservation followed by aggressive systemic chemotherapy. On November 17, 2020, the REI clinic initiated ovarian down-regulation with Leuprolide injections to protect the adnexa before chemotherapy. Starting November 23, 2020, the patient underwent four 21-day cycles of combination chemotherapy with Doxorubicin and Ifosfamide, completed by March 2021. She experienced manageable side effects. From June 2021 to mid-2023, she remained under six-month surveillance with regular CT scans confirming complete remission and resolution of preoperative hydronephrosis.

In late 2023, about three years after her initial surgery, routine pelvic CT scans revealed a new, hypervascular soft-tissue mass on the right pelvic side wall, with the patient remaining asymptomatic. The focus shifted from fertility preservation to life-saving local control. She underwent a re-exploratory laparotomy through her previous midline incision. Dense adhesions were lysed to access the pelvic retroperitoneum. Instead of a radical exenteration, a localized tumor excision was performed alongside a total abdominal hysterectomy (TAH) and bilateral salpingo-oophorectomy (BSO). The uterus and adnexa were removed as a single unit to prevent capsular breach. The previously repaired right internal iliac artery was carefully isolated, protected, and preserved during side wall dissection to avoid vascular injury.

Histopathology confirmed recurrent high-grade leiomyosarcoma with nuclear atypia and active mitosis, consistent with the 2020 tumor. The excision achieved clear margins (R0 resection), ensuring complete removal of the malignancy.

Post-hysterectomy management included close observation over chemotherapy due to an R0 resection and prior anthracycline exposure, reducing risks of bone marrow exhaustion and cardiotoxicity. Surgical menopause was managed with non-hormonal therapy to control vasomotor symptoms and prevent hormonal stimulation of mesenchymal pathways.

The patient underwent intensified surveillance, including clinical exams and cross-sectional imaging every 3 months for the first year, then every 6 months. Serial abdominal and pelvic CT scans through 2024, and May 2025, showed no evidence of disease, with a clear vaginal cuff and surgical bed and no soft-tissue masses. High-resolution chest CT scans and Doppler ultrasound confirmed the absence of metastases and preserved vascular and renal health. By May 2025, she was in sustained secondary complete remission with no evidence of disease.



Figure 1. Longitudinal computed tomography (CT) timeline of primary and recurrent high-grade right broad ligament leiomyosarcoma in a 28-year-old female patient.

Figure 1. Left: Sagittal baseline scan showing a giant 25 cm retroperitoneal pelvic-abdominal mass. Middle: Axial postoperative scan confirming vascular salvage and patency of the repaired right internal iliac artery (arrow). Right: Axial surveillance scan at three years revealing a localized hypervascular recurrence on the right pelvic sidewall (arrow).

CASE NO 2

The 64-year-old female patient presented with progressive abdominal distension and lower abdominal pain, leading to imaging surveillance. She has no known drug allergies or documented genetic abnormalities, including hereditary cancer syndromes such as leiomyosarcoma or renal cell cancer.

Approximately 8 months before presenting at King Khalid University Hospital, the patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy at a regional hospital in Najran. The surgical pathology report confirmed a primary diagnosis of highly aggressive spindle (conventional) broad ligament leiomyosarcoma, measuring at least 15 cm, and extracted and fragmented. No lymphovascular invasion was observed, and regional lymph nodes were not examined. Margins couldn't be definitively assessed due to tissue fragmentation. The initial postoperative plan involved close surveillance with regular pelvic contrast-enhanced MRI and CT scans of the chest, abdomen, and pelvis every 3 to 4 months.

The patient, referred to the Tertiary Gynecologic Oncology Unit for recurrent disease, showed a well-healed abdominal scar, excellent functional status (ECOG 1), stable vital signs, and obesity (BMI 34.3). Physical exam was unremarkable, with no tenderness or organomegaly. She was alert and maintained normal vital parameters.

Diagnostic assessment involved serial pelvic MRIs and contrast-enhanced CT scans to track the transition from postoperative fluid collections to a solid recurrence. A definitive scan revealed a 6.4 x 3.4 cm hypervascular pelvic mass invading surrounding structures. Surgical histopathology confirmed extensive intraperitoneal disease consistent with initial spindle cell morphology. Early imaging misrepresented the lesion as benign due to its loculated nature, but later development of a hypervascular mass with necrosis confirmed malignant recurrence. The diagnosis was recurrent, metastatic uterine leiomyosarcoma involving the pelvis, small bowel, cecum, and omentum. High-grade leiomyosarcomas are aggressive, with early recurrence indicating a high risk of systemic relapse requiring aggressive treatment.

On September 8, 2024, a specialized gynecologic oncology team performed a midline exploratory laparotomy, revealing severe intra-peritoneal adhesions, with small bowel adherent to the right anterior abdominal wall and the sigmoid colon fixed to the bladder. Multiple small peritoneal implants, cecal masses, separate ileal implants, an omental lesion, and a 6x4 cm ileal mass were identified. The procedure included extensive adhesiolysis, retroperitoneal dissection with bilateral ureter isolation, resection of the right pelvic/ileal mass, cecal masses, ileal implants, total omentectomy, and removal of an abdominal wall nodule. During bowel isolation, two small seromuscular injuries occurred more than 30 cm from the cecum, necessitating a 12 cm resection and primary anastomosis, verified leak-free via air and dye tests. Estimated blood loss was 500 mL; a pelvic drain was placed, and urine output remained adequate.

Perioperative management included 72 hours of IV metronidazole and cefuroxime, enoxaparin 40 mg daily for VTE prophylaxis, pantoprazole 40 mg BID, and multimodal analgesia with IV paracetamol and PCA, avoiding NSAIDs.

The pathology confirmed metastatic leiomyosarcoma in several specimens, including peritoneal mass, nodule, cecal mass, ileal implant, and abdominal wall lesion. The small bowel segment and omentum were negative for malignancy, with the latter showing benign tissue. The rectal implant was also benign.

Following complete surgical cytoreduction, the patient was referred to Medical Oncology on September 30, 2024, and started a systemic chemotherapy protocol due to the high risk of recurrence from multifocal intraperitoneal spindle cell sarcoma. The first-line regimen is Gemcitabine plus Docetaxel (GemTax), administered every 21 days with G-CSF prophylaxis. Alternative options for future progression include Doxorubicin monotherapy, limited by cardiac cumulative dose, or salvage therapies such as Trabectedin or Pazopanib, which target transcription pathways or tumor angiogenesis, respectively.

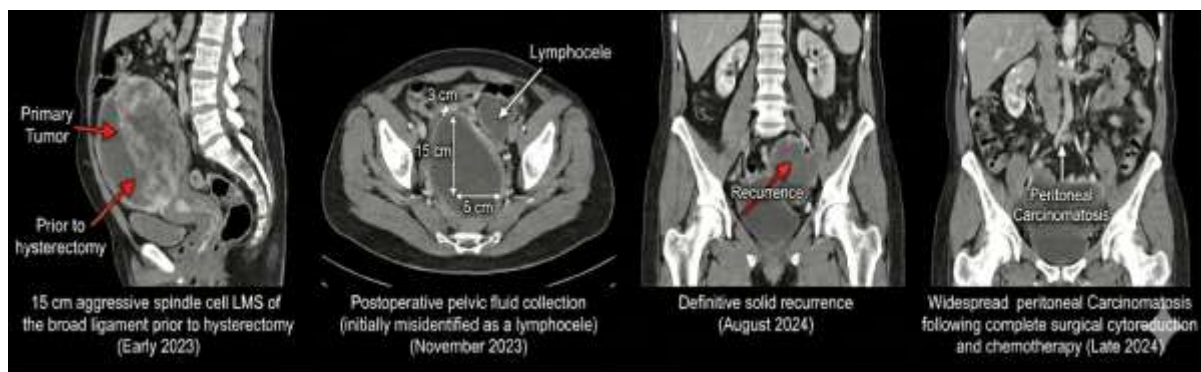


Figure 2. Serial computed tomography (CT) tracking of fragmented, recurrent, and metastatic broad ligament leiomyosarcoma transitioning from a suspected lymphocele to widespread peritoneal carcinomatosis.

Figure 2. Left: Baseline sagittal scan showing a 15 cm broad ligament mass before fragmented extraction. Middle-Left: Pelvic fluid collection initially tracked as a benign lymphocele. Middle-Right: Solid multi-visceral recurrence. Right: Coronal follow-up scan showing progression to extensive peritoneal carcinomatosis involving the ileum, cecum, and omentum

Table 1: Historical and Current Information From This Episode of Care Organized as a Timeline (Case Series of 2 Patients)

Clinical/Procedural Element	Case 1 (28-Year-Old Patient)	Case 2 (64-Year-Old Patient)
Primary Presentation & Symptoms	Late 2020: Progressive, severely painful pelvic-abdominal mass accompanied by rapid abdominal distension and severe right-sided hydroureter/hydronephrosis.	Early 2023: Progressive abdominal distension and lower abdominal pain. Treated at a regional hospital in Najran.
Primary Surgery	Late November 2020 (KKUH): Urgent exploratory laparotomy via an extended midline incision.	Mid-2023 (Regional Hospital): Primary Total Abdominal Hysterectomy and Bilateral Salpingo-Oophorectomy (TAH-BSO).
Primary Tumor Burden & Pathology	Giant 25 cm, encapsulated retroperitoneal mass originating from the right broad ligament (1913 gm, 22 X 17 X10 cm). High-grade spindle cell leiomyosarcoma with severe nuclear atypia, hyperchromatism, high mitotic rate, and focal lymphovascular invasion (LVI).	Massive tumor measuring at least 15 cm originating from the broad ligament. Aggressive spindle cell (conventional) leiomyosarcoma with active mitosis. No lymphovascular invasion noted.
Surgical Margins & Tissue Integrity	Complete macro-resection with negative microscopic margins, noted as a close margin of 0.1 cm. Tumor was excised fully intact.	Definite margins could not be pathologically assessed because the tumor mass was extracted and fragmented during the primary operation.
Primary Intraoperative Complications	Severe tumor adherence to the right pelvic sidewall resulted in an accidental laceration of the right internal iliac artery, causing temporary hemorrhagic shock.	None reported during the baseline procedure, though severe, dense intra-peritoneal adhesions developed post-operatively.
Acute Resuscitation & Interventions	The vascular surgery team achieved proximal control of the common iliac vessels and performed a successful primary suturing repair. Resuscitated with 3L blood loss replacement: 6 units PRBC, 6 units platelets, and 6 units FFP.	Standard postoperative recovery with no immediate hemorrhagic or vascular complications recorded.
Adjuvant Treatment & Fertility Care	Nov 2020 – Mar 2021: Referred to Reproductive Endocrinology (REI); initiated ovarian down-regulation with Leuprolide injections. Completed 4 cycles of combination Doxorubicin + Ifosfamide.	No immediate adjuvant chemotherapy was administered. Managed with standard radiological tracking every 3 to 4 months.
First-Line Remission & Tracking	June 2021 – Mid-2023: Complete clinical and radiologic remission under 6-month surveillance. Preoperative hydronephrosis fully resolved.	Nov 2023 – Apr 2024: Serial imaging monitored an operative bed fluid collection ($3 \times 2.5 \text{ cm}$), which regressed conservatively and was tracked as a benign lymphocele/seroma.
Recurrence Presentation & Timeline	Late 2023 (~3 years post-op): Routine follow-up pelvic CT revealed a new, asymptomatic hypervascular soft-tissue mass on the right pelvic side wall.	August 2024 (~15 months post-op): Pelvic MRI revealed a new 6 cm solid mass with central necrosis, growing rapidly to $6.4 \times 3.4 \text{ cm}$ within two weeks.
Recurrence Patterns	Localized Recurrence: Isolated mass confined to the right pelvic retroperitoneal side wall; no distant or intraperitoneal spread.	Metastatic Recurrence: Localized pelvic recurrence accompanied by widespread peritoneal carcinomatosis, ileal nodularities, and omental lesions.
Secondary Cytoreductive Surgery	Late 2023 (KKUH): Re-exploratory laparotomy with dense adhesiolysis, localized tumor excision, and a concurrent delayed TAH-BSO removed as a single unit.	September 8, 2024 (KKUH): Midline exploratory laparotomy, extensive adhesiolysis, bilateral ureter isolation, total omentectomy, mass resection, and abdominal wall nodule removal.

Secondary Organ & Vascular Handling	The previously repaired right internal iliac artery was carefully isolated, protected, and preserved to avoid vascular injury. Exenteration was successfully avoided.	The tumor was inseparable from the distal ileum. Bowel isolation caused two seromuscular injuries, requiring a 12 cm small bowel resection and leak-free primary anastomosis.
Recurrence Pathology & Margins	Confirmed recurrent high-grade leiomyosarcoma with active mitosis. Achieved a pathologically confirmed R0 resection (completely clear margins).	Confirmed high-grade metastatic leiomyosarcoma involving the pelvic peritoneum, cecum, ileum, and abdominal wall. Small bowel margins and omentum were benign.
Post-Recurrence Medical Oncology Plan	Managed with close clinical observation over chemotherapy due to the verified R0 resection margins and high risk of cumulative anthracycline cardiotoxicity.	September 30, 2024: Multidisciplinary Tumor Board recommended immediate systemic first-line chemotherapy with Gemcitabine + Docetaxel (GemTax) every 21 days + G-CSF.
Endocrine & Menopausal Management	Surgical menopause was managed strictly via non-hormonal therapy to prevent potential hormonal stimulation of mesenchymal pathways.	Patient was already postmenopausal at baseline presentation (64 years old); no hormone-replacement interventions indicated.
Long-Term Status & Follow-up	May 2025 Follow-up: Maintained on intensified 3-to-6 month surveillance. Serial CT scans and Doppler ultrasound confirm Sustained Secondary Complete Remission / No Evidence of Disease (NED).	Late 2024/2025 Follow-up: Referred out to Medical Oncology to undergo systemic therapeutic cycles; monitored via close post-surgical clinical oncology follow-up.

DISCUSSION

Strengths and Limitations

A key strength of our case series with two patients is the successful execution of aggressive, personalized multi-visceral secondary cytoreduction, achieving an optimal microscopic R0 resection in a challenging anatomical region. Despite intraoperative difficulties such as a life-threatening internal iliac artery laceration, a three-liter hemorrhage, and dense tumor adherence to vital organs, the multidisciplinary team effectively minimized morbidity through vascular salvage, anatomical preservation, and timely intestinal reconstruction. Additionally, our approach included advanced adjuvant strategies tailored to patient demographics, balancing fertility preservation with non-hormonal endocrine options to reduce risk.

A major limitation is the extremely limited clinical evidence for recurrent primary broad ligament leiomyosarcoma, a rare condition with few cases documented worldwide. Due to the lack of standardized treatment guidelines, our approach was based on general uterine sarcoma protocols. Moreover, an early pelvic recurrence was initially misinterpreted as a benign postoperative fluid, revealing a vulnerability in standard surveillance. This

Management practices for this rare tumor vary widely. Initial treatment typically involves excision of the mass, a total abdominal hysterectomy, and bilateral salpingo-oophorectomy. Pelvic lymph node dissection remains controversial and has not consistently demonstrated improved survival in uterine LMS. The role of adjuvant chemotherapy and radiotherapy is extrapolated from uterine LMS data, with conflicting results. Some studies suggest improved local control with radiotherapy, while others do not, often limited by small sample sizes. No definitive randomized trials exist for chemotherapy. In this case, adjuvant chemotherapy with single-agent doxorubicin was administered, given its tolerability and response in metastatic LMS [3-4].

Primary broad ligament leiomyosarcoma is an extremely rare mesenchymal cancer, with fewer than 50 cases documented in the English-language worldwide literature, as noted by Ahuja et al. [6]. Due to its rarity, developing standard surgical, radiotherapy, or chemotherapy protocols is particularly challenging, leading clinicians to adapt guidelines used for the uterus. Bouraoui et al. [7] found that early-stage, localized cases treated with definitive primary margin-negative surgery can result in short-term remission, with no recurrence observed after 15 months. Nonetheless, the overall outlook remains poor because of its aggressive local recurrence and typically late-stage presentations.

Metastatic leiomyosarcoma of the broad ligament is extremely rare, characterized by highly unusual and unpredictable spread patterns through both the bloodstream and lymphatic system. In addition to common sites like the lungs and liver, the literature reports remarkable visceral secondary metastases. Tian et al. (8) described late-occurring metastases to the pancreas and thigh following a 2.5-year disease-free period, which were

effectively managed with sequential radical metastasectomies. Likewise, Okagawa et al. (9) found gastric metastasis, and Cazzato et al. (10) documented a low-grade tumor spreading to the subcutaneous tissue of the neck through the Batson venous plexus before invading the myocardium as two intracardiac nodules.

Our conclusions are based on clinical outcomes from two contrasting cases. Case 1 shows that high-grade local recurrence can be effectively managed with meticulous, margin-negative secondary cytoreduction while preserving pelvic vasculature. In contrast, Case 2 illustrates that tissue fragmentation and delayed diagnosis of a recurrence as a benign lymphocele can lead to catastrophic peritoneal carcinomatosis. Therefore, aggressive margins, vigilant monitoring, and prompt chemotherapy are crucial to prevent rapid tumor progression.

Take-home message: Primary treatment of broad ligament leiomyosarcoma requires aggressive surgery with removal of the tumor and clear margins. Vigilant follow-up is crucial to detect early recurrence, especially in postoperative pelvic fluid. In case of recurrence or metastasis, secondary tumor removal combined with personalized systemic chemotherapy offers the best chance for long-term remission.

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