Journal of Obstetrics, Gynecology and Cancer Research | ISSN: 2476-5848

Advanced Leiomyosarcoma Treated with Total Abdominal Hysterectomy-Bilateral Salphingo Oophorectomy, Low Anterior Resection, and Bilateral Double J Stent: A Case Report

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Article Info

10.30699/jogcr.10.5.431

Received: 2025/01/05;
Accepted: 2025/03/01;
Published Online: 13 Mar 2025;

Use your device to scan and read the article online

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ABSTRACT

Uterine leiomyosarcoma (ULMS) is a rare neoplasm that accounts for only 1-2% of uterine malignancy. Despite its shallow occurrence, uterine sarcoma has a poor prognosis, which depends on the extent of the disease and its mitotic index. We report a case of 45-year-old woman with a 6-month history of massive uterine mass. On gynecology physical and vaginal examination, a palpable solid fixed mass 15 cm in size was observed in the lower abdomen and normal portio. A Multidisciplinary Team (MDT) including a gynecologic oncologist, urologist, gastrointestinal surgeon, radiologist, and anatomical pathologist decided for re-laparotomy, Total abdominal Hysterectomy-bilateral salphingo oophorectomy, low anterior resection, and protective loop-ileostomy. The final diagnosis was stadium 3A leiomyosarcoma. This case underscores the importance of early detection, comprehensive imaging, multidisciplinary approach, and continued exploration of innovative therapeutic strategies for managing this rare and aggressive tumor.

Keywords: Gynecology, Hysterectomy, Leiomyosarcoma, Oncology

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1. Introduction

Uterine leiomyosarcoma (ULMS) is a rare aggressive tumour with high recurrence rates, even when confined to the uterine corpus at the time of diagnosis (1). Although rare, the tumors are devastating with poor prognosis and aggressive biology leading to early metastatic spread both locally and distantly. Despite its shallow occurrence, uterine sarcoma has a poor prognosis, which depends on the extent of the disease and its mitotic index. With the initial diagnosis of metastatic disease, its 5-year survival rate is 10-15% (2). Preoperative diagnosis, pathology examination, and postoperative treatment are all challenging. This report presents a rare case of advanced ULMS exhibiting adhesion to the colon and rectum, which draws attention to the treatment with multidisciplinary approach therapy, as discussed by a multidisciplinary team (MDT), for advanced uterine leiomyosarcoma.

2. Case Presentation

A 45-year-old woman referred to the Dr. Soetomo General Academic Hospital in January 2024 with a 6-month history of massive uterine mass. On gynecology physical and vaginal examination, a palpable solid fixed mass 15 cm in size was observed in the lower abdomen and normal portio. MRI examination showed a solid mass with a cystic component with a partially indistinct border measuring approximately 11.7×12,25×12,4 cm in size, located posterior to the uterus, filling the pelvic cavity and adhered to the rectum and sigmoid colon with boundaries difficult to distinguish. The mass displays the uterus and bladder anteriorly and compresses both ureters, causing dilatation and moderate to severe ectasia of the bilateral pelvicocalyceal system. Laboratory studies showed a slight increase in tumor marker CA125 level of 43.6

u/ml, BUN level of 30 mg/dL, and Serum Creatinine level of 3.1 mg/dL Probable diagnoses of subserous myoma uteri, suspecting leiomyosarcoma, moderate to severe acute kidney injury, and moderate to severe bilateral hydronephrosis were made.

According to the diagnosis, the urologist performed a bilateral double J stent, and an open biopsy laparotomy was performed by the gastrointestinal surgeon. On the gross intraoperative examination, the retroperitoneal solid mass measuring 25 × 25 cm in size filled the pelvic cavity, and a normal-sized uterus with dense adhesion of the partial uterus with an anterior abdomen wall was observed (Figure 1). A histopathological examination was performed and revealed spindle mesenchymal cells with high mitotic activity (>20/10 HPF) and adherence to the muscularis externa of the colon and uterine serosa, supporting the diagnosis of leiomyosarcoma. IHC examination detected SMA and Desmic positive cytoplastic cell tumor, CD34 positive membrane cell tumor (Figure 2), S100 negative nucleus, and membrane cell tumor, DOG-1 negative cytoplastic cell tumor, CD117 negative membrane cell tumor with the conclusion of leiomyosarcoma. An MDT including a gynecologic oncologist, urologist, gastrointestinal surgeon, radiologist, and anatomical pathologist decided for re-laparotomy, Total abdominal Hysterectomy-bilateral salphingo oophorectomy, low anterior resection, and protective loop-ileostomy.

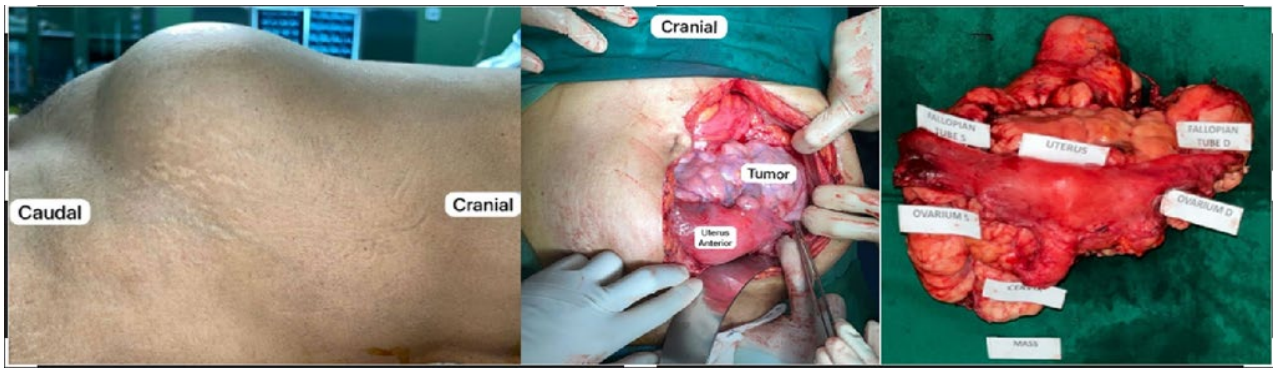


Figure 1. A normal-sized uterus with dense adhesion of the partial uterus with an anterior abdomen wall

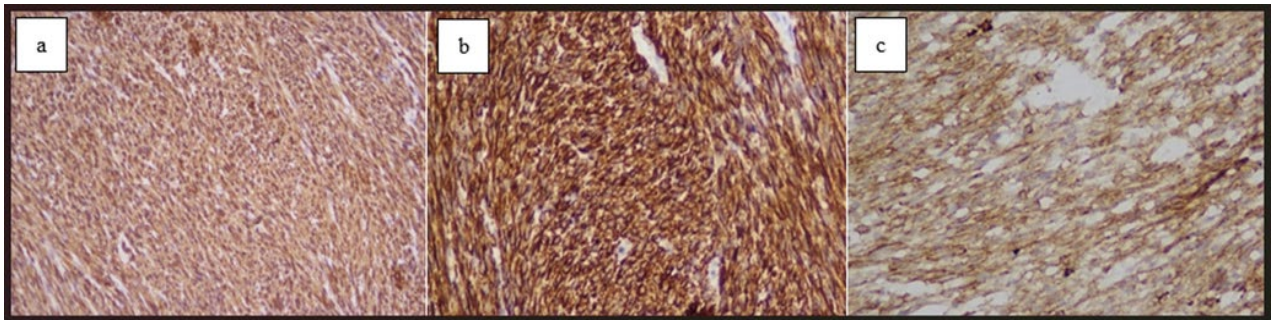


Figure 2. Spindle mesenchymal cells with high mitotic activity (>20/10 HPF) and adherence to the muscularis externa of the colon and uterine serosa

MDT joined the operation in March 2024. The finding was a solid mass with a hypervascular area adhered to the posterior and lateral uterus, which was 15 ×15 cm in size and adhered to mesocolon and mesosigmoid. The uterus volume was reduced postoperatively, and the mass was removed entirely. A postoperative pathological investigation suggested leiomyosarcoma measuring 20×15×12 cm in size, with no lymphatic invasion nor perineal invasion. The tumor was also grown in the muscularis externa colon, adhered to the uterus. There was carcinoma in situ cervix with glandular involvement located in the 5 and 6 o'clock position. Nevertheless, the final diagnosis was stadium 3A leiomyosarcoma. The tumor was removed entirely and the patient was planned to perform follow-up with physical examination and imaging once every 3 to 4 months for 2 to 3 years, continuing with once every 6-12 months due to mass complete resectability.

Routine follow up were done within 11 months after the surgery. There was no residual mass in the routine physical evaluation done every 3 months.

3. Discussion

This report presents a rare case of leiomyosarcoma involving adhesion to the colon and rectum. The diagnosis of leiomyosarcoma was confirmed through histopathological examination of postoperative specimens, which identified the tumor as stage IIIA according to International Federation of Gynecology and Obstetrics (FIGO) and American Joint Committee

on Cancer (AJCC) criteria, corresponding to pT3aNxMx in the TNM classification (3). No evidence of lymphatic invasion was observed, and tumor cells were confined to the muscularis externa of the colon, with adherence to the uterine serosa.

Leiomyosarcoma is a malignant neoplasm arising from the uterine wall's smooth muscle (4). The retroperitoneum, followed by the uterus, extremities, and trunk are the most common sites of leiomyosarcoma occurrence, as seen in the present case (5). Leiomyosarcoma accounts for 2% of uterine malignancy and 65% of uterine sarcoma (6). It is the most common subtype of uterine sarcoma, which also has the worst prognosis with the lowest survival rate (2) and its incidence increases with age. It occurs in the 3rd decade of life, but is most common in perimenopausal women or the 5th decade of life. Incidence at younger age is associated with genetic inheritance (5). Approximately, 6/1000 cases are diagnosed each year (2).

The clinical presentation of uterine leiomyosarcoma ranges from heavy and abnormal menstruation, anemia, pelvic pressure, and bulk symptoms such as urinary or bowel symptoms (2). Compression of the surrounding organs is frequently the cause of symptoms. Tumors can become very large before they cause symptoms in places like the retroperitoneum, as seen in our case (5). Although the majority (60%) of patients are detected in the early stages of the disease, more than 50% of cases relapse (7, 8). Despite being diagnosed in stage I, the overall 5-year survival rate is reported below 60% (8). Local recurrence and/or metastatic disease can still occur, especially in the lungs (4). An observational cohort study on 7455 patients with leiomyoma reported that survival was associated with the patient's age, higher comorbidity, black race, tumor's staging, grading, size, lymph node involvement, metastasis at diagnosis, positive surgical margin, adjuvant chemotherapy, and brachytherapy (9).

Pathological examination is required for the diagnosis with Stanford criteria, which is the most common method used to detect the presence of histologic atypia, tumor cell necrosis, and elevated mitotic rate. According to the Stanford criteria outlined, a histological diagnosis requires in the presence of at least two of the following: diffuse moderate-to-severe atypia, a mitotic rate of ≥10 figures per 10 HPF, or tumor necrosis (5, 10). Extrauterine locations require pretreatment biopsy; a core biopsy is the recommended method. After total resection, a detailed pathologic assessment is usually carried out (5). Histological analysis of the tumor revealed spindle mesenchymal cells resembling leiomyosarcoma, with tissue sections showing intersecting fascicles of proliferative oval to spindle-shaped cells, hyperchromatic nuclei, and elongated cytoplasm. Mitotic activity exceeded 20 mitoses per 10 high-power fields (HPF). Spindle-shaped cell morphology, high mitotic rates, and immunoreactivity for smooth muscle markers typically characterize spindle cell tumors. Variations in cell morphology, mitotic activity, and additional immunohistochemical markers highlight the heterogeneity of these tumors, emphasizing the balance between shared features and individual variability (11).

Immunohistochemistry distinguished leiomyosarcoma from other spindle cell tumors (12). Computed tomography (CT) scan or magnetic resonance imaging (MRI) can detect the lesion extension (13). Before the first suspicion, either by biopsy or myomectomy, expert pathologic review, molecular testing, imaging, and ER/PR testing can be considered. In the present case, the diagnosis was based on the patient's clinical presentation, imaging findings, and biopsy results. The abdomen and pelvis MRI showed a solid cystic mass with unclear borders and irregular margins. The mass, located posterior to the uterus, appeared isointense on T1-weighted imaging (T1WI) and iso- to hyperintense on T2-weighted imaging (T2WI). It filled the pelvic cavity with evidence of adhesion to the sigmoid colon and rectum. The mass also compressed both ureters, leading to moderate ectasia of the bilateral pelvicocalyceal systems. MRI provides detailed imaging without radiation, making it a valuable tool alongside PET or CT scans for evaluating potential metastasis when clinically indicated. Leiomyosarcoma typically appears on T1WI as a large, infiltrative myometrial mass with heterogeneous hypointensity and irregular margins. On T2WI, it demonstrates intermediate-to-high signal intensity, often with central hyperintensity indicative of extensive necrosis (7).

In clinical practice, individuals with leiomyosarcoma often require a multidisciplinary approach, treated with a combination of medicinal and surgical strategies (14). The primary treatment for early-stage leiomyosarcoma is hysterectomy with complete resection of visible tumor (15). The disease is divided into: limited to the uterus, known for suspected extrauterine disease, and disease that is not suitable for primary surgery. TAH-BSO is the surgical criteria for disease limited to the uterus and resectable extrauterine disease. On the other hand, systemic therapy, and/or EBRT and brachytherapy can be considered for diseases that are not suitable for primary surgery and non-resectable extrauterine disease due to not able to resection and/or not appropriate for surgery (16). Surgical approaches, including radical surgery, such as anterior resection, abdominoperineal resection, and local excision should aim to achieve macroscopically complete resection and microscopically minimal positive margins (9). In contrast to carcinomas, leiomyosarcoma has a low (5%–11%) lymph node involvement rate, and lymph node dissection is not required, as seen in the present case. An en-bloc resection with no tumor spillage should be attempted whenever the preoperative workup is worrying for uterine leiomyosarcoma. If extrauterine illness is curable, it should be removed. The diagnosis

is frequently obtained on an incidental myomectomy or hysterectomy material, which might need to be removed completely with a second-look procedure. However, the well-established treatment is early and complete surgical resection **(9)**. Adjuvant systemic therapies and radiotherapy have not shown significant benefits in improving the patients outcomes. Although no definitive treatments exist for advanced or recurrent leiomyosarcoma, recent advances have shown promise. Immunotherapeutic agents such as nivolumab and pembrolizumab, along with novel chemotherapeutics including olaratumab and pazopanib, have demonstrated potential efficacy in managing advanced cases **(17)**.

In the current case, the uterus was removed entirely, which was challenging, but successfully stopped the tumor cells from spreading. Based on the accurate analysis of clinicopathologic information and a review of the literature and guidelines, the appropriate management was established by several specialists. The patient free survival and overall survival rate were better than those documented in the literature by implementing a thorough management strategy.

4. Conclusion

This case report highlights the importance of the diagnostic and therapeutic challenges associated with this aggressive malignancy. The histopathological findings confirmed the diagnosis, revealing spindle-shaped mesenchymal cells with high mitotic activity, consistent with leiomyosarcoma. Imaging modalities, particularly MRI, played a crucial role in delineating the extent of the tumor and its impact on adjacent structures. This approach involves a team of specialists from various disciplines, including oncology, pathology, radiology, and surgery. Despite surgical intervention being the cornerstone for early-stage disease, treatment options for advanced or recurrent leiomyosarcoma remain limited. Emerging therapies, including immunotherapy and novel chemotherapeutic agents, can improve the patient outcomes, although further research is required to validate their efficacy. This case underscores the importance of early detection, comprehensive imaging, and continued exploration of innovative therapeutic strategies for managing this rare and aggressive tumor. A multidisciplinary approach is important because sarcomas are rare tumors that require complex diagnosis and imaging interpretation to achieve better patient outcomes.

5. Declarations

Acknowledgments

Not applicable.

Ethical Considerations

Our institution exempts ethical clearance for case reports. Patient consent for publication can be obtained by request to the author.

Authors' Contributions

AH contributed to patient care, conception of the case report, acquisition and interpretation of data, literature review, and drafting of the manuscript. HN and BAT contributed to the conception of the case report, literature review, and manuscript drafting. All authors approved the final manuscript.

Conflict of Interest

The authors declare no conflict of interest.

Fund or Financial Support

This research received no specific grant from any funding agency in the public, commercial, or not for profit sector.

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How to Cite This Article:

Herlambang, A., Nugroho, H., Tjokroprawiro, B. A. Advanced Leiomyosarcoma Treated with Total Abdominal Hysterectomy-Bilateral Salphingo Oophorectomy, Low Anterior Resection, and Bilateral Double J Stent: A Case Report. J Obstet Gynecol Cancer Res. 2025;10(5):431-5.

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