



Received: 07-11-2025
Accepted: 17-12-2025

International Journal of Advanced Multidisciplinary Research and Studies

ISSN: 2583-049X

Surgical Management of Advanced Endometrial Stromal Sarcoma: A Case Report

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Abstract

Endometrial stromal sarcoma (ESS) is a rare uterine malignancy characterized by nonspecific clinical presentation and aggressive biological behavior, particularly in high-grade forms. Due to its rarity, optimal management remains challenging, with surgery representing the mainstay of treatment. We report the case of a 66-year-old woman with a history of gastrointestinal stromal tumor who was diagnosed with high-grade endometrial stromal sarcoma. The patient initially underwent intestinal reconstruction with termino-terminal anastomosis and bilateral adnexectomy. During subsequent surgery, a large tumor of uterine origin

with invasion of the urinary bladder was identified. Complete en bloc resection of the tumor, including regional mesenteric lymph nodes, was achieved. Histopathological examination confirmed high-grade ESS (AJCC TNM 2017: pT1b Nx Mx). The postoperative course was uneventful, and the patient was referred for adjuvant chemotherapy. This case demonstrates that radical en bloc surgical resection is feasible even in advanced high-grade ESS. Comprehensive surgical management remains crucial for achieving optimal disease control, reducing recurrence risk, and improving long-term outcomes.

Keywords: Endometrial Stromal Sarcoma, High-Grade, En Bloc Resection, Surgical Management

Introduction

Endometrial stromal sarcoma (ESS) is a rare and aggressive malignant neoplasm originating from the stromal cells of the endometrium. According to the literature, ESS accounts for less than 0.2% of all uterine tumors, placing it among the rarest gynecological malignancies. The disease most commonly affects women in middle and older age groups, with a mean age ranging between 42 and 58 years, although cases in younger women have also been reported (Puliyath & Nair, 2012) [1].

Clinical presentation is often nonspecific, which complicates early detection and diagnosis. Patients frequently present with abnormal uterine bleeding or symptoms resembling benign conditions such as uterine fibroids or endometriosis, underscoring the importance of thorough clinical, radiological, and histopathological evaluation. ESS is classified into low-grade and high-grade subtypes, with high-grade tumors typically exhibiting a more aggressive clinical course, a higher propensity for metastasis, and increased recurrence rates.

Historically, treatment of ESS has relied primarily on surgical ablation. Currently, complete surgical resection is considered the cornerstone and most critical modality of treatment, with additional therapeutic options determined by disease stage and histopathological characteristics (Capozzi *et al.*, 2020) [2].

Case report

We present the case of a 66-year-old woman with a medical history significant for a gastrointestinal stromal tumor (GIST), who underwent intestinal reconstruction with a termino-terminal anastomosis and bilateral adnexectomy in February 2024.

Histopathological examination confirmed that the primary tumor was an endometrial stromal sarcoma. Following the initial surgical procedure, the patient was referred for adjuvant oncological treatment by the gynecologic oncology multidisciplinary team. During the second surgical intervention, a large tumor of primary uterine origin was identified, with infiltration of the urinary bladder. The tumor was completely excised, including resection of the regional mesenteric lymph nodes. Histopathological analysis confirmed a high-grade endometrial stromal sarcoma (AJCC TNM 2017: pT1b Nx Mx). The patient experienced an uneventful postoperative recovery during hospitalization and was subsequently referred for adjuvant chemotherapy after discharge.

Discussion

Endometrial stromal sarcoma (ESS) is a rare but aggressive neoplasm that requires a high level of surgical expertise due to its potential for local invasion, recurrence, and distant metastasis. The primary treatment modality for both low-grade and high-grade ESS remains complete surgical excision, which typically involves hysterectomy with or without resection of adjacent structures, depending on the extent and stage of the disease.

Laparoscopic and robotic-assisted approaches may be employed in selected cases; however, open laparotomy is often preferred for large or complex tumors. Surgical management of ESS should aim to be as radical as possible, with complete tumor removal in a single specimen (en bloc resection) to prevent tumor cell dissemination and reduce the risk of recurrence. Minimal tumor manipulation prior to complete resection is strongly recommended, particularly in high-grade and bulky tumors, in accordance with the “tumor containment” principle (Viossat *et al.*, 2021) [4].

Controversy persists regarding the necessity of resecting adjacent organs such as the urinary bladder, rectum, or major blood vessels in cases of local invasion or metastasis. International clinical guidelines support aggressive radical surgery in cases with extensive invasion or suspected metastatic spread (Gadducci *et al.*, 2021) [5]. Combined surgical strategies utilizing both open and minimally invasive techniques, including laparoscopic and robotic-assisted procedures, are increasingly applied in smaller tumors or selected clinical scenarios to reduce perioperative morbidity and enhance postoperative recovery. Nevertheless, open surgery remains the preferred approach for large tumors and extensive disease due to superior exposure and a higher likelihood of achieving complete resection.

Key challenges in the surgical management of ESS include the risk of tumor cell dissemination during manipulation, the difficulty of achieving complete resection in advanced-stage disease, and the potential for recurrence even after macroscopically complete excision (Tajiri *et al.*, 2021) [6]. Potential complications include injury to the urinary bladder, rectum, major blood vessels, and neural structures; therefore, surgical management often necessitates a multidisciplinary operative team and multiple intraoperative procedures. Emerging evidence highlights the importance of a combined treatment approach, integrating surgical resection with systemic therapies, including hormonal and targeted treatments, particularly in high-risk or metastatic disease (Gadducci *et al.*, 2023) [3].

Conclusion

This case demonstrates that en bloc tumor resection may be feasible even in high-risk forms of endometrial stromal sarcoma, thereby improving treatment outcomes and aligning with recent studies emphasizing the pivotal role of surgery in enhancing survival and minimizing recurrence (Capozzi *et al.*, 2020) [2]. Comprehensive and meticulous surgical management, particularly en bloc resection, significantly improves the likelihood of successful disease control in high-grade ESS and may have a substantial impact on long-term patient outcomes.

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