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Report

Subserosal Myoma Associated with Stump (Smooth Muscle Tumors of Uncertain Malignancy Potential): A Case Report

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Abstract

Smooth muscle tumors of the uterus are typically benign, but a rare subset known as smooth muscle tumors of uncertain malignant potential (STUMP) poses diagnostic challenges. First classified by the World Health Organization in 2003 due to their atypical histological features, STUMPs are difficult to distinguish from benign leiomyomas and malignant leiomyosarcomas. This case study describes a 53-year-old woman with a large abdomino-pelvic mass, initially suspected to be sarcoma. Clinical and imaging evaluations, including MRI, led to exploratory laparotomy, revealing a subserosal myoma associated with STUMP. Histopathological analysis confirmed the diagnosis, underscoring the complexities in differentiating STUMPs from other uterine tumors. STUMPs account for a portion of uterine sarcomas, and accurate diagnosis relies on specific histological criteria, including nuclear atypia, mitosis index, and tumor necrosis. Advanced imaging, such as dynamic MRI, helps improve differentiation between STUMP, leiomyosarcoma, and leiomyoma. Although STUMPs are rare, they typically have a better prognosis than leiomyosarcomas, with lower recurrence rates. However, due to their uncertain behavior, patients require long-term monitoring. The limited understanding of STUMPs highlights the need for continued research and clinical vigilance to improve diagnosis and management strategies.

Keywords

STUMP, Leiomyomas, Leiomyosarcomas

1. Introduction

Smooth muscle tumors of the uterus are common and mostly benign. Some tumors may present unusual anato-mo-pathological aspects, leading to problems of differential diagnosis, notably with leiomyosarcoma. In 2003, the World Health Organisation classified these tumours as smooth muscle tumours of uncertain malignant potential (STUMP) [1].

These are complex tumors, whose histological characteristics do not allow them to be classified as either benign or malignant [2]. Only clinical follow-up will reveal the true

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nature (benign or malignant) of the tumor. Numerous studies have sought to identify diagnostic markers for these tumors, based on histological and immunohistochemical criteria, but to date, none of these studies have defined the category of STUMP [3]. The absence of consistent diagnostic criteria and the uncertainties surrounding the diagnosis of STUMP have led to its overdiagnosis over the years [4]. In fact, according to Ip et al., a diagnosis of STUMP is appropriate when a tumor presents any unusual combination of the three previously mentioned features but does not meet the Stanford criteria for leiomyosarcoma. [5, 6]

This complexity in diagnosis underscores the importance of detailed case reports to enhance clinical understanding, as demonstrated in the following case study.

2. Case Presentation

We present the case of a 53-year-old female patient, without any particular personal history or family history of cancer, single, nulliparous nulligest. She was referred by her primary care physician to the National Institute of Oncology in Rabat, Morocco, for an abdomino-pelvic mass. The patient initially presented with altered general condition associated with constipation and pollakiuria.

Our clinical examination on admission revealed a patient in good general condition: conjunctiva was normally colored, blood pressure and heart rate were normal, BMI = 30, WHO = 0, ASA = 1.

Palpation of the abdomen revealed a large (25 cm long axis), hard, difficult to mobilize abdomino-pelvic mass.

Gynecological examination revealed a healthy cervix, and abdominal palpation combined with vaginal palpation revealed a mobile mass adjacent to the uterus. Rectal palpation revealed no infiltration or mass. The rest of the examination was normal.

Pelvic MRI revealed:

- An abdomino-pelvic mass of uterine origin measuring 19 x 17.5 x 25 cm, initially suggestive of sarcoma.
- A 76x62x175 mm left abdomino-pelvic formation, shaping the vascular structures, extending retroperitoneally and shaping the left ovarian vein. No pelvic adenopathy and small pelvic effusion.
- A 9 cm cystic lymphangioma of the left ovarian vein reaching the left renal vein.

As part of the investigations, a thoracoabdominal CT scan was performed which showed the absence of distant metastases but the presence of peritoneal nodules associated with sarcomatosis.

After multidisciplinary consultation, the patient underwent exploratory laparotomy in March 2022 and was found to have a uterine mass adherent to the small intestine, mild ascites, and nodules of peritoneal carcinosis: Total colpohysterectomy with bilateral adnexectomy, with resection of 80 cm of the last loop of bowel adherent to the mass, and with terminal-terminal bowel-gastric anastomosis. Partial resection of the left cystic lymphangioma was also performed. Anatomopathology revealed: A Total weight of 6525g. Very large subserosal myoma measuring 30 x 22 x 17cm associated with a smooth muscle tumor of uncertain malignant potential (STUMP).

Non-tumoral ileal resection, Lymph node biopsy: 01N-/01N,

Cytology: inflammatory ascitic fluid, Non-specific acute ulcerated appendicitis. No histological signs of malignancy.

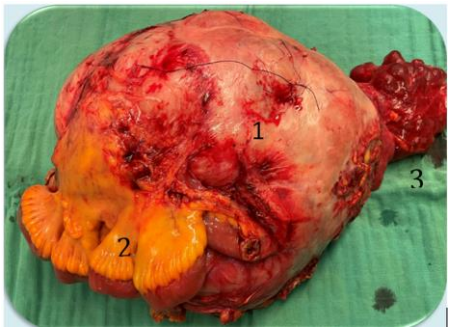


Figure 1. *Surgical specimen: 1-Polymyomatous uterus, 2- Grecic loop, 3-Cystic lymphangioma.*

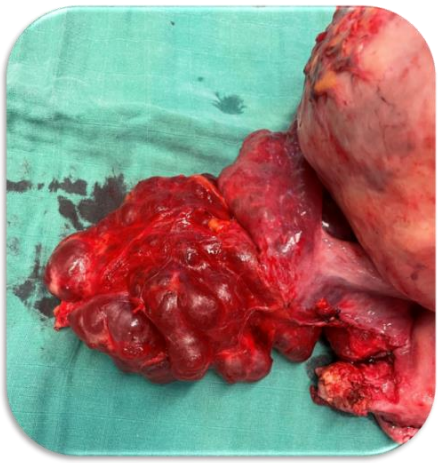
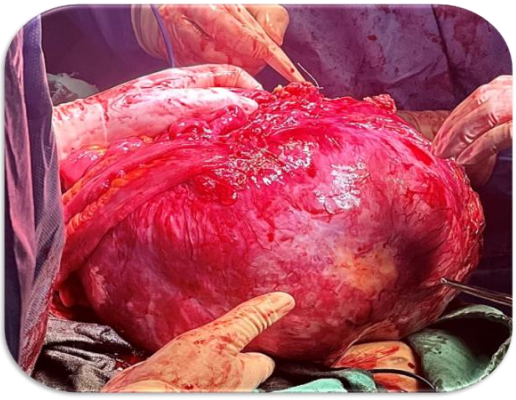


Figure 2. *Surgical specimen: better visualization of cystic lymphangioma.*



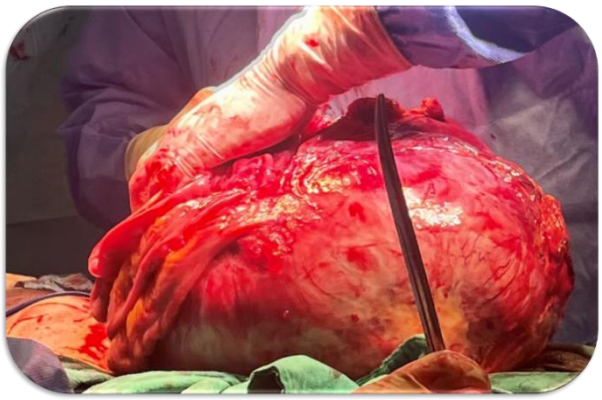


Figure 3. *Two peroperative images of abdomino-pelvic mass resection with adhesiolysis.*

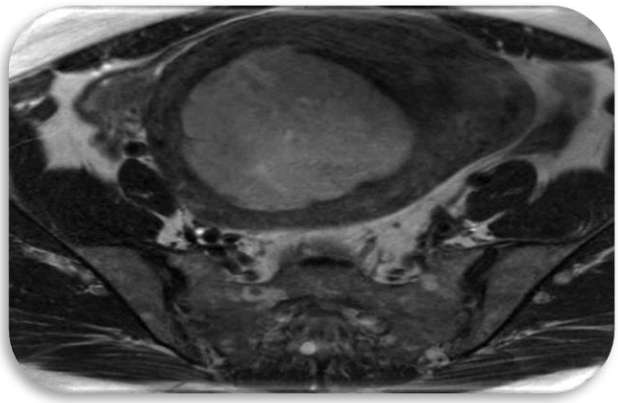


Figure 4. *Scan image of a smooth muscle tumor of uncertain malignant potential of the uterus.*

Because of moderate signal intensity on T2-weighted scans, mass was thought to be smooth muscle tumor of uncertain malignant potential [10].

3. Discussion

There are numerous uterine smooth muscle tumours with well-defined clinical outcomes and histological criteria; These tumors are challenging to classify due to their overlapping features with both benign leiomyomas (LM) and malignant leiomyosarcomas (LMS), making definitive diagnosis difficult. Between these two extremes lie several variants with unusual histological features, clinical course and prognosis.

These are smooth muscle tumors of uncertain malignant potential [1]. STUMPs are very rare tumours, accounting for 1/3 of uterine sarcomas and 1.3% of uterine cancers [2].

Diagnosis of smooth muscle tumors is challenging due to a lack of specific features [1], and it is clinically indistinguishable from leiomyoma (LM) with no imaging techniques to differentiate them [2]. However, contrast-enhanced dynamic MRI can distinguish between LMS, STUMP, and LM with 94% accuracy and 96% specificity [7]. Proton emission tomography with 18F Fluorodeoxyglucose (18F-FDG) has also shown high uptake in this tumor [8]. A thoraco-abdomino-pelvic CT scan should be requested to check for metastases, particularly pulmonary metastases from STUMP [1].

Histologically, three criteria are used to classify smooth muscle tumors as benign or malignant: nuclear atypia, mitosis index, and presence of tumor necrosis. Two of these criteria are required for a malignancy diagnosis. STUMP occurs when one malignancy criterion is present, but the second is difficult to assess [1]. Currently, no antibody can definitively identify the malignant nature of a smooth muscle tumor, although P53 and P16 have shown promise in uterine LMS, STUMP, and LM [9].

The CGH array technique can detect small chromosomal abnormalities not visible on standard karyotyping, aiding STUMP classification. If a STUMP has a flat chromosomal profile, it may suggest a leiomyoma, while abnormalities commonly seen in leiomyosarcomas suggest closer monitoring. However, the tumor's morphology remains the key factor in diagnosis, not its genomic profile [9].

Thus, the therapeutic approach differs from one school to another, ranging from simple myomectomy to total hysterectomy or even bilateral adnexectomy [1]. The preferred treatment for recurrence is surgical excision, followed by adjuvant therapies such as pelvic irradiation, chemotherapy (including doxorubicin and cisplatin), medroxyprogesterone, and gonadotropin-releasing hormone analogues [11-14].

The prognosis for STUMP is better than for LMS. Various studies have shown a significantly lower recurrence rate than LMS, and 5-year survival rates ranging from 92% to 100%. Given the uncertain nature and evolution of these tumors, patients should receive close, long-term clinical and radiological follow-up [1].

As noted by Ip et al., STUMPs are associated with the potential for delayed recurrences. However, they exhibit a higher median survival rate following recurrence compared to more aggressive malignant uterine neoplasms [15-17].

4. Conclusion

In conclusion, STUMPs are a rare and still poorly understood group of smooth muscle tumors. They are difficult to diagnose, as their clinical and radiological features are the same as those of leiomyomas, so only anatomopathology enables the diagnosis to be made. The prognosis is still uncertain, but studies suggest that it is better than for leiomyosarcomas.

Abbreviations

STUMP	Smooth Muscle Tumors of Uncertain Malignant Potential
MRI	Magnetic Resonance Imaging
BMI	Body Mass Index
WHO	World Health Organization
ASA	American Society of Anesthesiologists
LM	Leiomyomas
LMS	Leiomyosarcomas

Author Contributions

Meryem Lamrani: Conceptualization, Investigation, Resources, Writing – original draft, Writing – review & editing
Khaoula Lakhdar: Supervision
Oumaima Sardaoui: Validation
Yacir Alami: Methodology, Supervision, Validation
Fouad Tijami: Supervision, Validation

Conflicts of Interest

The authors declare no conflicts of interest.

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