Insights into Management of Smooth Muscle Tumours with Uncertain Malignant Potential: A Case Series

AMEY CHUGH¹, JANVI PATEL², VARSHINI VADITHALA³, PRASHANSA GUPTA⁴

(CC) BY-NC-ND

ABSTRACT

Obstetrics and Gynaecology Section

Smooth Muscle Tumours with Uncertain Malignant Potential (STUMPs) represent a rare and diagnostically challenging category within uterine neoplasms. These tumours exhibit biological features similar to both benign leiomyomas and malignant leiomyosarcomas, complicating diagnosis and management. STUMPs are characterised by their potential for unpredictable clinical behaviour, including local recurrence and, in rare cases, metastasis, necessitating careful postoperative monitoring. Classifying uterine mesenchymal tumours is difficult due to significant overlap with terms such as atypical leiomyoma, atypical leiomyoma with low risk of recurrence, and atypical leiomyoma with low malignant potential. Despite their infrequent occurrence, STUMPs have garnered increasing attention due to their ambiguous nature and the clinical implications they pose. Diagnostic criteria and management strategies for STUMPs remain areas of active research and debate within gynaecological pathology and oncology. The present case series was aimed to contribute to the existing body of literature by presenting three cases of female patients (aged 38-year-old, 34-year-old and 47-year-old) of large uterine leiomyomas that were later diagnosed as STUMPs postoperatively following hysterectomy. Each case underscores the complexities involved in diagnosing and managing these tumours, emphasising the importance of multidisciplinary collaboration and long-term follow-up in optimising patient outcomes.

Keywords: Atypical leiomyoma, Diagnostic challenges, Hysterectomy, Myomectomy, Postoperative monitoring, Uterine leiomyoma, Uterine neoplasms

INTRODUCTION

The STUMPs represent a rare and diagnostically challenging category within uterine neoplasms. These tumours exhibit biological features similar to those of benign leiomyomas and malignant leiomyosarcomas, posing difficulties in both diagnosis and management [1]. STUMPs are characterised by their potential for unpredictable clinical behaviour, including local recurrence and, in rare cases, metastasis, necessitating careful postoperative monitoring and management decisions [2,3]. Due to the significant overlap in terms such as STUMP, atypical leiomyoma, atypical leiomyoma with low risk of recurrence and atypical leiomyoma with low malignant potential, classifying uterine mesenchymal tumours is difficult [4].

Despite their infrequent occurrence, STUMPs have garnered increasing attention due to their ambiguous nature and the clinical implications they pose. The diagnostic criteria and management strategies for STUMPs remain areas of active research and debate within the fields of gynaecological pathology and oncology.

CASE SERIES

Case 1

A 38-year-old Para 2 Living 2 (P2L2) woman with a history of tubectomy eight years back presented with an irregular menstrual cycle for two years and heavy menstrual bleeding with clots, along with abdominal pain for the past two months, without any pressure symptoms. She reported a history of regular menses until two years ago. The general examination was normal; however, the abdominal examination revealed a mass of 22-24 weeks in size, which was mobile side-to-side. On per vaginal examination, the uterus felt 22-24 weeks in size, with left forniceal fullness.

The ultrasound showed a uterus measuring $14.7 \times 8 \times 10.5$ cm, with a large intramural fibroid on the posterior left lateral wall measuring $10.6 \times 9 \times 10$ cm. This fibroid had peripheral vascularity, minimal internal vascularity, and an endometrial thickness of 11 mm. The ovaries were normal in size and morphology. Blood investigations,

including Complete Blood Count (CBC), Liver Function Tests (LFT), Renal Function Tests (RFT), Glycated Haemoglobin (HbA1C), etc., were within normal limits.

The patient underwent a hysterectomy with a left salpingooophorectomy due to the size of the fibroid and the associated symptoms. Histopathology confirmed the presence of a STUMP with tissue necrosis only (measuring 14.5×10×8 cm and weighing 900 g), in addition to non specific cervicitis, an unremarkable left fallopian tube, and a simple follicular cyst of the left ovary [Table/Fig-1].

Case 2

A 34-year-old P2L2 tubectomised woman (who underwent laparoscopic tubal ligation 10 years ago) presented with complaints of urinary incontinence and an increase in abdominal girth for the past 1.5 months. She reported no abdominal pain, vaginal bleeding, or discharge, and her bowel movements were normal. Her menstrual cycles were regular, and a general examination was unremarkable.

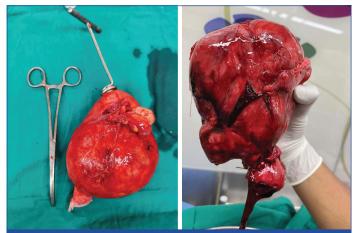
On abdominal examination, a midline firm mass measuring 24-26 weeks was noted, which was mobile laterally and non tender. On per vaginal examination, a mass attached to the anterior wall of the uterus, measuring 16-18 weeks in size, was found. The uterus was not separately palpable but was freely mobile, with clear and non tender bilateral fornices.

Ultrasonography revealed a retroverted uterus with no focal myometrial lesions and an endometrial thickness of 7 mm. A large, lobulated solid mass with multiple fibrous septae in the lower abdomen, reaching up to the umbilicus and measuring 18×8.5×17 cm, raised suspicion of ovarian dysgerminoma due to internal vascularity. The mass compressed the uterus, resulting in the loss of fat planes posteriorly.

An Magnetic Resonance Imaging (MRI) of the abdomen and pelvis with contrast revealed a large lesion (10.4×14.8×16.4 cm) likely arising from the uterus (fibroid), which was inseparable from the anterior and left lateral uterine walls. A suspicious bridging sign was noted, along with compression of the uterus and left ovary

posteriorly, as well as, compression of the bilateral iliac vessels and ureters, causing bilateral hydronephrosis and hydroureter.

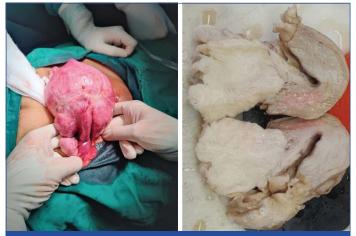
Based on the examination findings, a total abdominal hysterectomy was planned. Histopathological examination confirmed a STUMP weighing 1.3 kg and measuring 15×14×8 cm, with an elevated mitotic index but without cellular atypia or tissue necrosis [Table/Fig-2].



[Table/Fig-1]: Specimen of uterus (with myoma) with cervix, left fallopian tube and ovary of size 14.5×10×8 cm (Case 1). **[Table/Fig-2]:** Specimen of uterus (with myoma) of size 15×14×8 cm (Case 2). (Images from left to right)

Case 3

A 47-year-old P2L2 woman with a history of two caesarean deliveries and a tubectomy which was done twenty years back was experiencing heavy menstrual bleeding with clots for six months and abdominal pain for the past 4-5 months, with no urinary or bowel disturbances. The general examination was normal. The abdominal examination revealed a soft and non tender abdomen, while the per vaginal examination indicated a uterus of 10 weeks in size, with free and non tender bilateral fornices. The ultrasound showed a uterus measuring 10×9.5×7 cm with multiple fibroids and an endometrial thickness of 13 mm. Both ovaries were normal in size and morphology. Laboratory investigations were within normal limits. The patient underwent a total abdominal hysterectomy [Table/Fig-3]. The cut section revealed a uterus with STUMP [Table/Fig-4]. Histopathology confirmed the presence of a STUMP measuring 7×8×8 cm and weighing 700 g, with cellular atypia, without tissue necrosis or elevated mitotic index.



[Table/Fig-3]: Intraoperative picture of uterus (with myoma) (Case 3). **[Table/Fig-4]:** Gross specimen of the uterus with STUMP (Case 3). (Images from left to right)

DISCUSSION

Kempson used the term "STUMP" for the first time in 1973 [5]. STUMPs still pose significant diagnostic challenges due to their overlapping features with benign leiomyomas and malignant leiomyosarcomas [6]. According to the World Health Organisation (WHO), any uterine smooth muscle tumour that is not classified as benign or malignant falls into the STUMP category [1,5]. The heterogeneity within STUMPs necessitates a thorough histopathological examination to distinguish them from other uterine smooth muscle tumours. The criteria for diagnosing STUMPs are not standardised and can include apical mitotic figures, moderate to severe cytological atypia, and a low mitotic index [1]. In the present series, the histopathological examination post-hysterectomy was crucial in confirming the diagnosis, highlighting the need for meticulous pathological evaluation.

Clinical presentations of STUMPs are diverse, ranging from asymptomatic incidental findings to symptomatic presentations that mimic benign uterine fibroids or malignant tumours. Symptoms commonly reported include abnormal uterine bleeding, pelvic pain, urinary symptoms due to compression, and abdominal distension [7]. For instance, cases have been documented where STUMPs were mistaken for large fibroids or ovarian tumours based on imaging findings, necessitating careful pathological evaluation postsurgery [3,5,6,8,9]. Tumour borders and their relationships with the surrounding myometrium and cellularity represent additional morphological criteria in the diagnosis of STUMP. Histologically, STUMP is diagnosed by a mitotic count of 10 high-power fields equal to or less than 10, coagulative necrosis, and none-to-mild atypia [10]. The markers of STUMP include cystic structures, non uniformity, ill-defined borders, mixed echogenicity, moderate to rich inner vascularisation, and the absence of fan-shaped shadowing. Overdiagnosis of this neoplasia has occurred due to diagnostic uncertainties and a lack of uniform diagnostic criteria for STUMP [11].

Surgical resection remains the cornerstone of treatment for STUMPs, typically involving hysterectomy or myomectomy, depending on factors such as tumour size, patient age, fertility desires and clinical symptoms [3,8,9]. The choice between conservative management and aggressive surgical intervention is often influenced by the lack of clear guidelines on the behaviour and optimal management of STUMPs [3,8].

The patients in the present series belong to the age group of 34-47 years old. Two patients out of three had complaints of heavy menstrual bleeding along with abdominal pain, while one of them had only pressure symptoms with increasing abdominal girth. One out of the three cases showed a raised suspicion of ovarian dysgerminoma on ultrasound, which was diagnosed as a fibroid compressing the bilateral ureters on Magnetic Resonance Imaging (MRI). All three patients underwent surgical management (hysterectomy), and microscopically, all three showed histopathological findings of STUMP.

The case series presented by Dall'Asta A et al., included patients ranging from 44-51 years of age, similar to the cases we presented. The subjects in the present study did not undergo any adjuvant therapy, and no recurrences were observed [1]. A previously published case series reported three cases, one of which developed recurrence with evidence of diffuse lung metastases nine years after hysterectomy, but the patient was clinically stable [12].

The case presented by Akad F et al., involved a 50-year-old post-menopausal female who complained of abdominal pain, an enlarged abdomen, and a constant sensation of abdominal pressure. MRI showed the presence of a voluminous left ovarian tumour. Peroperatively, a large deformed uterus was observed, and microscopically, the mitotic index was elevated, but there was no clear cellular atypia or necrosis, suggesting that while the tumours were atypical, they were not definitively malignant [3].

As this is a rare entity, there is insufficient demographic data to consolidate the hypothesis based on the age of occurrence. There is no standard protocol approved for the treatment of patients suspected of having STUMP. The management of choice is hysterectomy, although myomectomy can also be performed if fertility preservation is desired.

Amey Chugh et al., Case Series of STUMP: Insights into Diagnosis and Management

CONCLUSION(S)

The management of STUMPs remains an area of ongoing debate, with surgical resection being the mainstay of treatment. Hysterectomy is often recommended, particularly when the tumour size, symptoms and imaging characteristics suggest a potential for malignancy. However, in cases where fertility preservation is a priority, myomectomy can be considered. The lack of standardised diagnostic criteria and treatment guidelines necessitates a personalised approach to each case, involving a multidisciplinary team of gynaecologists, pathologists and radiologists. Long-term follow-up is crucial due to the risk of local recurrence and, in rare cases, metastasis. The authors experience with these three cases emphasises the need for continued research and collaboration in the field of gynaecological pathology and oncology to improve diagnostic accuracy and optimise management strategies for patients with STUMPs.

REFERENCES

- [1] Dall'Asta A, Gizzo S, Musarò A, Quaranta M, Noventa M, Migliavacca C, et al. Uterine smooth muscle tumours of uncertain malignant potential (STUMP): Pathology, follow-up and recurrence. Int J Clin Exp Pathol. 2014;7(11):8136-42.
- Di Giuseppe J, Grelloni C, Giuliani L, Delli Carpini G, Giannella L, Ciavattini [2] A. Recurrence of uterine smooth muscle tumour of uncertain malignant potential: A systematic review of the literature. Cancers (Basel) [Internet]. 2022;14(9):2323.

- [3] Akad F, Filip B, Mocanu V, Akad M, Acatrinei C, Scripcariu V. Rare case of smooth muscle tumour of uncertain malignant potential- clinical case. Maedica (Buchar) [Internet]. 2021;16(2):302-06.
- [4] Tinelli A, D'Oria O, Civino E, Morciano A, Hashmi AA, Baldini GM, et al. Smooth muscle tumour of uncertain malignant potential (STUMP): A comprehensive multidisciplinary update. Medicina (Kaunas) [Internet]. 2023;59(8):1371.
- [5] Gadducci A, Zannoni GF. Uterine smooth muscle tumours of unknown malignant potential: A challenging question. Gynecol Oncol [Internet]. 2019;154(3):631-37.
- [6] Lin YM, Hong SY, Teng SW, Chang CK, Lai TJ. Retrospective analysis on characteristics of uterine smooth muscle tumours of uncertain malignant potential-13 years' experience. Clin Exp Obstet Gynecol [Internet]. 2022;49(10):234.
- Ip PP, Tse KY, Tam KF. Uterine smooth muscle tumours other than the ordinary [7] leiomyomas and leiomyosarcomas: A review of selected variants with emphasis on recent advances and unusual morphology that may cause concern for malignancy. Adv Anat Pathol [Internet]. 2010;17(2):91-112.
- Hughes L, Roex A, Parange A. STUMP, a surprise finding in a large fibroid [8] uterus in a 20-year-old woman. Int J Womens Health [Internet]. 2018;10:211-14.
- [9] Vahedpoor Z, Khamechian T, Zandi N. Uterine Smooth Muscle Tumors of Uncertain Malignant Potential (STUMP) mistaken with ovarian tumor in a female with polio: A case report. J Obstet Gynecol Cancer Res. 2017;2(1):01-03. Doi: 10.5812/ogcr.9425.
- [10] Bell SW, Kempson RL, Hendrickson MR. Problematic uterine smooth muscle neoplasms. A clinicopathologic study of 213 cases. Am J Surg Pathol. 1994;18(6):535-58.
- [11] Ip PPC, Cheung ANY, Clement PB. Uterine smooth muscle tumours of uncertain malignant potential (STUMP): A clinicopathologic analysis of 16 cases. Am J Surg Pathol [Internet], 2009:33(7):992-1005.
- Berretta R, Rolla M, Merisio C, Giordano G, Nardelli GB. Uterine smooth muscle [12] tumor of uncertain malignant potential: A three-case report. Int J Gynecol Cancer. 2008;18(5):1121-26.

PARTICULARS OF CONTRIBUTORS:

- Assistant Professor, Department of Obstetrics and Gynaecology, Dr. D. Y. Patil Medical College, Hospital and Research Centre, Pune, Maharashtra, India.
- Resident, Department of Obstetrics and Gynaecology, Dr. D. Y. Patil Medical College, Hospital and Research Centre, Pune, Maharashtra, India. 2.
- Resident, Department of Obstetrics and Gynaecology, Dr. D. Y. Patil Medical College, Hospital and Research Centre, Pune, Maharashtra, India. З.
- Resident, Department of Obstetrics and Gynaecology, Dr. D. Y. Patil Medical College, Hospital and Research Centre, Pune, Maharashtra, India. 4.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR: Dr. Janvi Patel.

B8, Carnation Girls Hostel, Dr. D. Y. Patil Medical College, Sant Tukaram Nagar, Pune-411018, Maharashtra, India, E-mail: 97janvipatel@gmail.com

AUTHOR DECLARATION:

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: Aug 24, 2024
- Manual Googling: Nov 28, 2024
- iThenticate Software: Nov 30, 2024 (11%)

Date of Submission: Aug 23, 2024 Date of Peer Review: Oct 25, 2024 Date of Acceptance: Dec 02, 2024 Date of Publishing: Feb 01, 2025

ETYMOLOGY: Author Origin **EMENDATIONS:** 6