The Journal of Obstetrics and Gynecology of India (November–December 2022) 72(6):536–538
https://doi.org/10.1007/s13224-021-01560-4

CASE REPORT

Leiomyosarcoma Occurring After Hysterectomy for Benign Fibroids: A Case Report

Kaustubh Girish Burde[1] · Anupama Rajanbabu[2] · Indu R. Nair[3] · Pavithran Keechilat[4]

Received: 23 June 2021 / Accepted: 19 August 2021 / Published online: 31 January 2022

Keywords Leiomyosarcoma · Leiomyoma · Hysterectomy · Recurrence

Abbreviations

STUMP	Smooth Muscle Tumour of Uncertain Malignant Potential
FDA	Food and Drug Administration
LDH	Lactate Dehydrogenase
CA 125	Cancer Antigen 125
Hpf	High power field
SMA	Smooth Muscle Actin

Introduction

Uterine sarcomas are highly malignant tumours accounting for less than 1% of gynaecologic malignancies and 2–5% of all uterine malignancies in women. Leiomyosarcoma is defined as malignant smooth muscle tumour displaying spindle cell morphology. In 2014, according to the FDA, the prevalence of unsuspected uterine leiomyosarcoma in patients undergoing hysterectomy or myomectomy for benign pathology was 1 in 498.

Case Report

A 50-year-old lady presented with complaints of abdominal swelling for 1 year. She gave history of abdominal hysterectomy with bilateral salpingo-oophorectomy done three years ago. The histopathology report of the specimen reported that the specimen was received in multiple pieces which measured 30 × 40 × 6 cm when put together. The slide and block review was reported as leiomyoma with infarctoid necrosis and focal bizarre nuclei. No evidence of malignancy was seen (Fig. 1 and Fig. 2).

The computed tomography scan showed a solid lobulated nodular lesion measuring 10 × 11 cm in the abdominopelvic region extending from rectovesical pouch into the peritoneum with peritoneal deposits. Her tumour markers were normal. (LDH—261 and CA 125—25.9).

On laparotomy, a large bosselated mass of 12 × 10 cm was present in lower abdomen adherent with sigmoid colon, vaginal vault, and bladder. There was also a round firm deposit of 4 × 5 cm infiltrating into para-vesical space with dense adhesion to bowel loop superiorly and ureter inferiorly. Multiple seedling lesions (3 × 4 cm) were seen in left paracolic space, retroperitoneal space and inferior surface of the liver. Complete cytoreduction was achieved.

The resected tumour consisted of lobulated grey-white mass measuring 12 × 9 × 7 cm. The cut surfaces showed multiple lobulations, with whirling. Microscopy showed diffuse pleomorphism, atypical mitosis and tumour

Dr.Kaustubh Girish Burde is a M.Ch. Resident, Amrita Institute of Medical Sciences, Kochi, Kerala, India. Dr.Anupama Rajanbabu is a Professor and HOD, Department of Gynaecology Oncology, Amrita Institute of Medical Sciences, Kochi, Kerala. Dr. Indu R. Nair is a Professor,Department of Pathology, Amrita Institute of Medical Sciences, Kochi, Kerala. Dr. Pavithran Keechilat is a Professor and HOD, Department of medical Oncology, Amrita Institute of Medical Sciences, Kochi, Kerala.

✉ Kaustubh Girish Burde
kaustubhburde@yahoo.com

Anupama Rajanbabu
anupamashyam@gmail.com

Indu R. Nair
drinduharikrishnan@gmail.com

Pavithran Keechilat
pavithrank@aims.amrita.edu

[1] Amrita Institute of Medical Sciences, Kochi 682041, Kerala, India

[2] Dept. of Gynaecology Oncology, Amrita Institute of Medical Sciences, Kochi, Kerala, India

[3] Dept of Pathology, Amrita Institute of Medical Sciences, Kochi, Kerala, India

[4] Dept of Medical Oncology, Amrita Institute of Medical Sciences, Kochi, Kerala, India

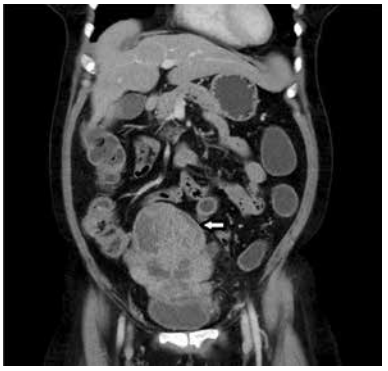

Fig. 1 Solid lobulated nodular lesion measuring 11 × 10 cm in the abdominopelvic region extending from rectovaginal pouch into the peritoneum

necrosis. Mitosis was noted (1–2)/ hpf including atypical mitosis. In view of these findings, a diagnosis of leiomyosarcoma was made. Immunohistochemistry showed the tumour cells to be positive for SMA, h-caldesmon, p53 and negative for desmin, thus confirming the diagnosis. Slide review of initial uterine specimen showed stromal tumour with infarctoid necrosis and focal atypia suggestive of STUMP.

After discussion in multidisciplinary tumour board, she was started on chemotherapy. She underwent four cycles of gemcitabine–docetaxel. Post-four cycles, reassessment CT scan showed an ill-defined enhancing sheet of soft tissue in retro-vesical pouch along left lateral wall of urinary bladder and multiple well-defined lesions in both lobes of liver. The case was discussed in multidisciplinary tumour board again. She was started on second-line chemotherapy (adriamycin + Ifosfamide). After six cycles, the patient remains asymptomatic.

Discussion

According to the latest World Health Organization definition, uterine STUMPs are smooth muscle tumours with features that preclude an unequivocal diagnosis of leiomyosarcoma, neither do they fulfil the criteria for leiomyoma, nor its variants, thus raising concern that the neoplasm may behave in a malignant fashion. It was found that among women undergoing hysterectomy or myomectomy for a presumed diagnosis of leiomyoma, 0.01% were diagnosed as STUMP. The recurrence rate after hysterectomy for a STUMP is 7.3%. The mean disease-free interval is 42 months. The presence of leiomyosarcoma after STUMP is very rare.

Andrea Dall'Asta et al. reported five cases of uterine masses treated by surgical procedure diagnosed as STUMP on final pathology. All patients in the study remained without recurrence with a follow-up period ranging from 6 to 81 months [1].

Antonio Maccio described a case report of a patient, who had undergone myomectomy for a suspected uterine myoma histologically proven to be STUMP, who came with diffuse abdominal sarcomatosis. She underwent surgery achieving a complete cytoreduction. Post-op histological examination showed high-grade uterine leiomyosarcoma. She showed relapse of leiomyosarcoma in the peri-gastric region for which she underwent complete removal of the neoplastic recurrence. She remained disease-free at the last follow-up [2].

Tsukasa et al. published a report of a 52-year-old woman who was diagnosed to have submucosal fibroids and had undergone total laparoscopic hysterectomy 4 years before presenting with a leiomyosarcoma. She underwent laparotomy, and the pathological evaluation confirmed the tumour to be a leiomyosarcoma. A review of the initial tissue did not show presence of any malignancy [3].

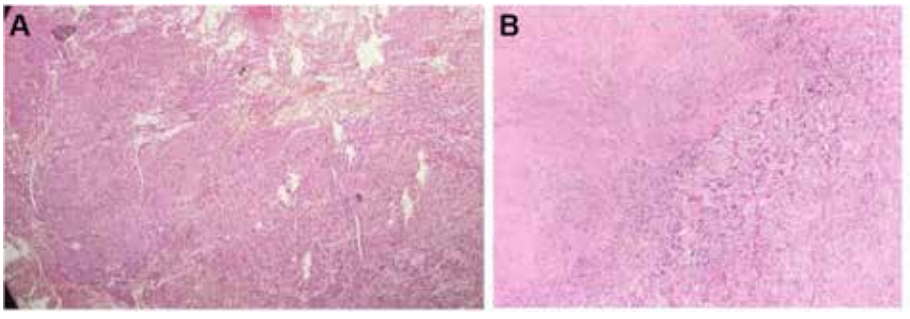

Fig. 2 **a** Cellular neoplasm composed of cells arranged in fascicles, bundles and sheets. Neoplastic cells were spindly with elongated to ovoid vesicular nuclei, few showing condensed chromatin. Scattered bizzare cells noted with high to moderate nuclear pleomorphism, irregular vesicular nuclei, condensed chromatin and prominent nucleoli. Mitosis was noted 1-2/hfp including atypical mitosis. Large areas of tumour necrosis was noted, surrounded by the acellular areas. Focal areas of hyalinisation was noted in between. Microscopy was suugestive of leiomyosarcoma **b** Microscopic picture showed stromal tumour with infarctoid necrosis and focal atypia suggestive of stromal tumour of unknown malignancy potential (STUMP)

Jasmine Tan-Kim et al. reported a case of leiomyosarcoma developing in previously hysterectomized lady with benign pathology. Uterus was removed by morcellation. Recurrence occurred 6 years after initial surgery [4].

The lady in this case report is 50 years of age with BMI of 30. Her tumour markers were within normal range. The initial histopathology report was suggestive of a benign tumour. She underwent open surgery without use of morcellator, but the hysterectomy specimen was removed in pieces according to that pathology reports. There is a gap of three years between the initial hysterectomy and current mass which is less than the median time to first recurrence after initial diagnosis of STUMP.

What is unique in this case is that there is presence of leiomyosarcoma in the patient after three years of an open hysterectomy showing STUMP pathology. Ndo lung nodules or distant metastasis were noted. This could be de novo synthesis of leiomyosarcoma within peritoneal cavity without a uterus or fibroid being present in situ.

Conclusion

This is a unique case showing leiomyosarcoma arising 3 years after the primary hysterectomy for fibroid uterus diagnosed on retrospective analysis as STUMP. Therefore, any case of secondary solid abdominal mass occurring after the initial surgery for benign conditions may have a chance of being leiomyosarcoma and should be evaluated in detail.

Author Contributions All authors contributed to the patient management and follow-up.

Funding There were no funds or grants involved.

Declaration

Conflict of interest None of the authors have any potential conflict of interest.

Ethical Statement Informed consent was obtained from the patient for presenting her data for publication. All parts of Declaration of Helsinki have been applied.

Informed consent The patient involved in this case was appropriately consented for this publication using the institution's policy for publication.

References

1. Dall'Asta A, Gizzo S, Musarò A, et al. Uterine smooth muscle tumors of uncertain malignant potential (STUMP): Pathology, follow-up and recurrence. Int J Clin Exp Pathol. 2014; 7(11):8136–42. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4270570/
2. Macciò A, Chiappe G, Kotsonis P, et al. Abdominal leiomyosarcomatosis after surgery with external morcellation for occult smooth muscle tumors of uncertain malignant potential: A case report. Int J Surg Case Rep [Internet]. 2017; 38:107–10. DOI: https://doi.org/10.1016/j.ijscr.2017.07.020 https://www.sciencedirect.com/science/article/pii/S2210261217303395?via%3Dihub
3. Takahashi T, Ugajin T, Imai N, et al. Leiomyosarcoma after Total Laparoscopic Hysterectomy with Power Morcellation. Case Rep Obstet Gynecol. 2019; 2019:1–4. https://pubmed.ncbi.nlm.nih.gov/31662931/
4. Tan-Kim J, Hartzell KA, Reinsch CS, et al. Uterine sarcomas and parasitic myomas after laparoscopic hysterectomy with power morcellation. Am J Obstet Gynecol. 2015;212(5):594.e1-10. https://doi.org/10.1016/j.ajog.2014.12.002 (**Epub 2014 Dec 11 PMID: 25499259**).

About the Author

Dr. Kaustubh Girish Burde, is a second year M.Ch. student in the Department of Gynaecology Oncology in Amrita Institute of Health Sciences, Kochi. He has completed his DGO from Pramukhswami medical college, Karamsad, Gujarat and DNB (OBGY) from Narayana Hridayalaya, Bengaluru, Karnataka