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Which Behavior of Myomatous masses is Diagnostic for Leiomyosarcoma: How Do You Clinically Differentiate Between Leiomyoma and Leiomyosarcoma? A Case Report

Leila Mousavi Seresht[1], Amir Reza Farhadian Dehkordi[2], Azar Danesh Shahraki[1]*, Fereshteh Mohammadizadeh[3], Fedyeh Haghollahi[4]

1. *Department of Obstetrics and Gynecology, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran*
2. *Research Student, Isfahan University of Medical Sciences, Isfahan, Iran*
3. *Department of Pathology, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran*
4. *Vali-E-Asr Reproductive Health Research Center, Family Health Research Institute, Tehran University of Medical Sciences, Tehran, Iran*

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Corresponding Information:
Azar Danesh Shahraki,
Department of Obstetrics and Gynecology, School of Medicine, Isfahan University of Medical Sciences, Isfahan, Iran

Email: Danesh@med.mui.ac.ir

ABSTRACT

Distinguishing between leiomyosarcoma and benign uterine leiomyoma, as one of the common problems in reproductive age, brings a great challenge in dealing with patients. Clinical findings, imaging report, tumor markers, endometrial biopsy, and even age of the patients will not be helpful to make this distinction. So, the important question is when we should consider leiomyosarcoma probability? In this report, an unexpected case of leiomyosarcoma with a poor prognosis is presented. A 45-year-old female patient gravida 3 with complaint of hypermenorrhea from one year ago was presented to the gynecologic department. The patient did not mention anything special in her medical history. At last, a hysterectomy was planned due to her irreversible bothering symptom. The final histologic report was leiomyosarcoma in the smallest myomatous mass of the uterus, and her post-operation CT scan with contrast of abdomen-pelvic revealed several metastatic focuses. Uterine sarcomas as one of the most important causes of mortality worldwide have a low incidence and defining the standard treatment and the exact prognosis require further study. More definitive ways to differentiate benign uterine leiomyoma from leiomyosarcoma including genetic studies, specific imaging techniques, specific tumor markers, and investigating the method of intrauterine biopsy by flexible or robotic hysteroscopy are needed.

Keywords: Leiomyoma, Leiomyosarcoma, Abnormal Uterine Bleeding, Myomectomy, Hysterectomy

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Introduction

Uterine smooth muscle tumors consist of three groups, benign masses or leiomyomas, smooth muscle tumor of uncertain malignant potential (STUMP), and malignant ones or leiomyosarcomas. Leiomyoma is one of the most prevalent pathologies in reproductive ages, which includes more than 80% of females in this age **(1)**. However, uterine sarcomas with aggressive nature and high mortality potential consist of only 1-3% of uterine malignancies that are mostly reported in postmenopausal ages **(2-4)**. The prognosis for uterine leiomyosarcoma is poor. The 5-year overall survival rate in uterine leiomyosarcoma is around 19–65% in all stages, and 52–85% in women with stage I–II disease **(5)**.

Differentiating between uterine leiomyoma and leiomyosarcoma is still challenging, neither the rapid growth of uterine mass, nor vaginal bleeding and pelvic pain are in favor of malignancy **(3)**. Moreover, what makes it more complicated is the weakness of diagnostic methods such as magnetic resonance imaging or endometrial biopsy in detecting these pathologies **(3, 4)**. Considering the fatal behavior of leiomyosarcoma with a maximum of 42% survival rate after 5 years in comparison with the benign feature of leiomyoma in females of reproductive age with no need for surgical treatment on most occasions, differentiating these two pathologies is of great concern. In the present study, a pre-menopausal patient

with a surprising diagnosis of leiomyosarcoma is reported.

Case Presentation

A 45-year-old gravida3 para3, a previously healthy woman, was referred with a history of abnormal uterine bleeding since 6-month earlier. The only finding in her physical examination was the enlarged uterus that later was defined to be related to multiple leiomyomas. According to the patient's age and the duration of her complaint, endometrial sampling was recommended. Although the endometrial sampling report was benign and in favor of proliferative phase, the patient insisted on performing a hysterectomy due to her long-lasting bleeding. An abdominal hysterectomy with preservation of ovaries was planned. The histological report of the uterine specimen was surprising; there were multiple intramural benign leiomyomas and one well-defined mass measuring 2.8 cm with moderate to severe atypia and high mitotic activity (more than 10 mitoses/10HPF) without necrosis in the fundus; in other words, a leiomyosarcoma (Figure 1). The other surprising point was the illumination of multiple sites of metastasis in pelvic computerized tomography (CT) scan less than one month from primary surgery, which was confirmed by needle biopsy (Figure 2).

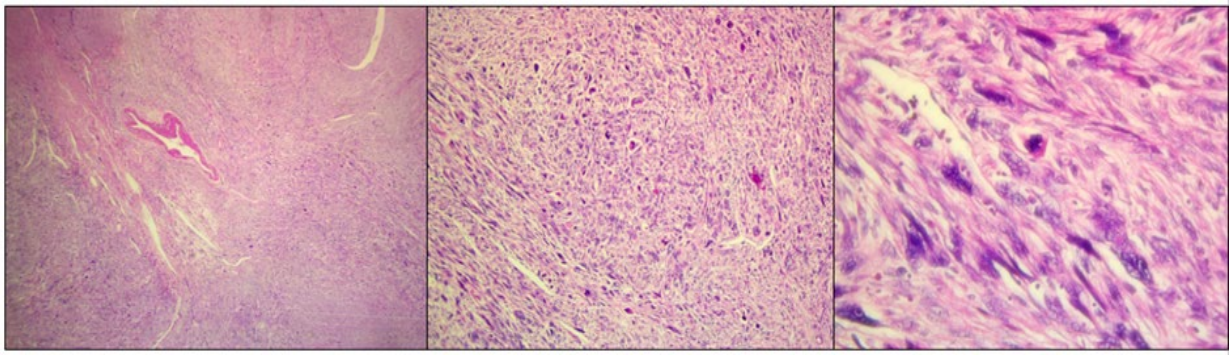


Figure 1. Microscopic examination of the malignant mass of the uterus revealed multifocal moderate to severe atypia with up to 12 mitoses at 10 HPF without signs of necrosis (H & E staining; from left to right: x40, x100, and x400 magnification)



Figure 2. Pelvic CT scan showed multiple sites of pelvic metastases shortly after hysterectomy

Discussion

Leiomyosarcoma, with a prevalence of 1-2.5 per 100,000 women, has mostly been reported in patients over 50 years old **(2)**. Based on the literature, the probability of leiomyosarcoma diagnosis is estimated at approximately 16% in a patient who had been undergone surgery for expected uterine leiomyoma less than 7 cm, but the chance is much less at a younger age, like the present case, which is about 0.9% **(2, 4)**. To take into consideration the importance of leiomyosarcoma co-incidence in patients with pre-surgical suspicion of leiomyoma, and lower chance of survival in case of misdiagnosis within laparoscopic morcellation and malignant cells dissemination, the FDA emphasized on much higher probability of this diagnosis, in about 1 per 498 cases of leiomyoma **(1, 2)**. Distinguishing between these two pathologies

remains challenging for clinicians, as neither clinical symptoms nor ultrasonographic findings are determined **(6)**. Due to the limited efficacy of the available methods, mentioning patients' risk factors like black race, long-term use of tamoxifen, or previous pelvic radiotherapy and in some cases, the presence of necrosis or ill-defined margin in pelvic MRI could be helpful. However, as in present case, the patient history is not remarkable and there is remaining debate on the best candidate for surgery. There is not enough evidence on the safety of diagnostic needle biopsy in the case of suspicious mass, the fans of this method defended its efficacy in differentiating malignancy in the retroperitoneum, but others criticize this modality in spillage of malignant cells **(7-9)**. Although based on the available evidence, there is a low but possible chance of malignant cell seeding even via vaginal, image-guided, or minimally invasive laparoscopic biopsy.

An investigational study on the intrauterine approach by robotic devices is suggested, which could eliminate the risk of intraperitoneal or vaginal metastasis in case of malignancy **(8)**. The occurrence of pelvic recurrences in less than one month from primary surgery in the present case supported the hypothesis of malignant cells dissemination in non-experienced hands even within open surgery, which significantly decrease the patient survival **(10, 11)**. It is worthy to note that the reported case in the present study had shown a dramatic response to external beam radiotherapy plus chemotherapy and had not experienced any more episodes of recurrences after one year. A point that should be explained by the difference in genetic translocation within the different tumors, it is the time to investigate more on the rule of molecular heterogenicity or supplemental agent on prognosis.

Conclusion

Uterine sarcoma, as one of the fatal gynecological malignancies, is one of the most worrisome issues in the management of leiomyoma, especially in patients at reproductive age. There is a need for further study on differentiating these two distinct pathologies, including sampling methods like robotic surgery, genetic evaluation, tumor markers, and so on.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Authors' Contributions

All authors read and approved the final manuscript.

L.MS: designed the work. AD.SH: designed the work and drafted the manuscript. AR.D and FM: edited the manuscript and interpreted the data. L.MS: interpreted the data. F.M and F.H: edited the manuscript.

Ethical approval

This study was performed according to the Helsinki statements, and the personal data of the patients did not appear for anyone. Informed consent was received from the patient for using her data.

Conflict of Interest

The authors reported no conflict of interest.

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