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Medical Journal of Obstetrics and Gynecology

Case Report

A Case of Uterine Leiomyosarcoma Presenting as Hemoptysis

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Submitted: 08 May 2023

Accepted: 07 June 2023

Published: 10 June 2023

ISSN: 2333-6439

Keywords

- Uterine leiomyosarcoma
- Fibroids
- Hemoptysis

Abstract

Uterine leiomyosarcoma is a rare malignancy with a known tendency for pulmonary metastases. The diagnosis and treatment of this condition can be challenging due to the rarity of disease. The authors present the case of a 48-year-old female with a history of uterine fibroids who presented with hemoptysis and was found to have pulmonary metastases due to uterine leiomyosarcoma. We discuss the challenges in diagnosis and review the current guidelines on surveillance for uterine masses.

INTRODUCTION

Uterine leiomyosarcoma is a rare and aggressive malignancy with poor prognosis. Most cases are discovered at an early stage, but recurrence rates are high, varying from 45 to 75%. The most common site of disease recurrence is the lung [1]. This condition is difficult to diagnose because there are currently no serum markers, clinical features, or imaging modalities that can reliably distinguish a benign uterine fibroid from a malignant leiomyosarcoma [2,3]. For patients who experience menstrual irregularities, anemia, pelvic pain or pressure, or obstructive symptoms, hysterectomy can be offered as a definitive solution. The incidence of unexpected sarcomas discovered from hysterectomies done for benign indications is about 0.1-0.6% [4]. The only way to definitively diagnose uterine sarcoma is surgically but there are less invasive ways to manage fibroids such as medical therapies like hormonal treatments or uterine artery embolization which may be preferred by younger patients or older patients with medical contraindications to surgery [2]. In this case report, we review the clinical course of a patient with a longstanding history of presumed fibroids who developed hemoptysis and was found to have uterine leiomyosarcoma metastatic to her lungs. The aim of this report was to review ways in which fibroids can be followed so that early malignant disease can be detected prior to spread to distant sites.

CASE PRESENTATION

A 35-year-old nulligravid female and Jehovah' s Witness with past medical history of uterine fibroids, obesity with body mass index 34, and type II diabetes mellitus was referred to obstetrics and gynecology for abnormal uterine bleeding. Initial transvaginal pelvic ultrasound showed multiple right ovarian cysts but no evidence of uterine lesions. Repeat surveillance ultrasound done several months later showed a new submucosal fibroid in the left lateral aspect of the uterus measuring 0.8 x 0.9 x 0.9 cm. She then began experiencing dysmenorrhea, dyspareunia, and menorrhagia and had surveillance pelvic ultrasounds done roughly every three to six months in the first few years then every two to four years. These serial ultrasounds showed multiplying and enlarging fibroids. The largest fibroid on the most recent pelvic ultrasound measured 11.03 x 12.53 x 10.01 cm. Hysterectomy versus uterine artery embolization for the management of her menorrhagia were discussed with the patient and she opted to proceed with bilateral uterine artery embolization. The risk of morbidity and mortality from hysterectomy was thought to be high since she is a Jehovah's Witness and refused blood products.

A few months later at age 48, she presented to urgent care with hemoptysis. Her chest X-ray showed multiple bilateral pulmonary masses. A computed tomography (CT) scan of her chest, abdomen and pelvis was obtained and revealed innumerable pulmonary parenchymal and pleural-based nodules and masses concerning for metastatic disease. There was no evidence of disease in the abdomen or pelvis aside from her known uterine masses. An ensuing respiratory infectious workup was negative (Figure 1).

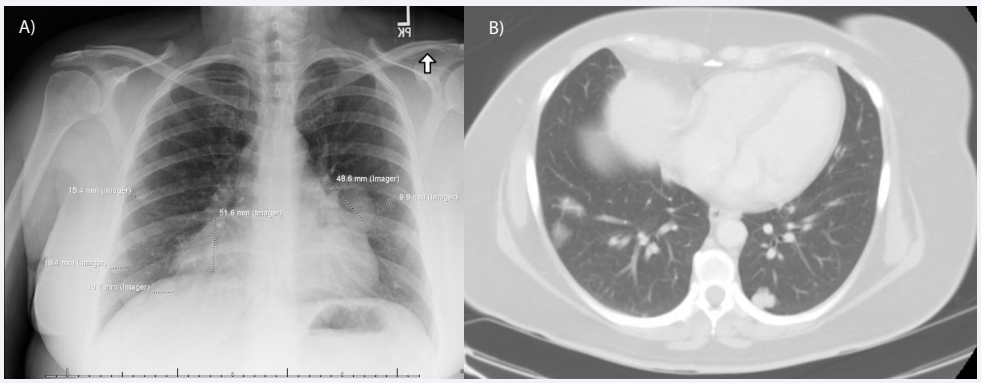

Figure 1 A) X-ray and B) CT of the chest showing multiple bilateral pulmonary masses.

She underwent flexible fiberoptic bronchoscopy with bronchoalveolar lavage which showed bleeding from the lingula without endobronchial lesions visualized. The cytology from transbronchial brushing of the lingula was unrevealing (Figure 2).

Given her history of uterine fibroids, extrauterine leiomyoma or leiomyosarcoma involving the lung was suspected. Positron emission tomography (PET) scan of the whole body was performed and showed intense radiotracer uptake in a left uterine wall mass and right adnexa, as well as among multiple pulmonary nodules including a hemorrhagic right lower lobe nodule (Figure 3).

Ultimately the patient underwent CT-guided transthoracic needle biopsy of a right pleural mass that was positive for metastatic high-grade sarcoma with smooth muscle differentiation. The tumor cells were positive for smooth muscle actin (SMA) and negative for desmin and pancytokeratin which is consistent with leiomyosarcoma [5] (Figure 4).

The patient was referred to gynecologic oncology and she was started on systemic chemotherapy with gemcitabine and docetaxel due to metastatic disease. After starting treatment, she developed worsening hemoptysis requiring hospital admission and bronchial artery embolization. Repeat CT scan of her chest done during this admission showed enlarging pulmonary nodules. She continued to decline clinically despite multiple cycles of chemotherapy and ultimately decided to transition to comfort care (Figure 5).

DISCUSSION

We reviewed this case to evaluate how this patient's fibroids could have been followed to allow for earlier detection. According to the American Cancer Society, the 5-year survival is 62% for localized disease, 34% for regional disease, and 13% for distant disease [6]. Screening for uterine malignancies has not been feasible and there are currently no evidence-based guidelines for the surveillance of fibroids [7,8]. An annual office visit and pelvic ultrasound at the minimum has been suggested by the American

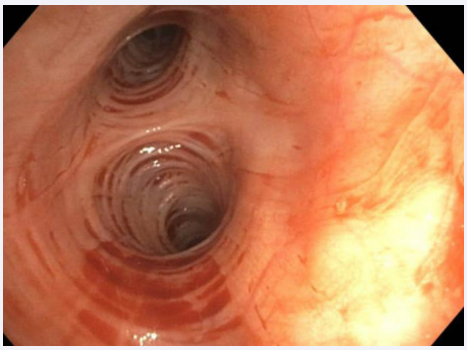

Figure 2 View of left bronchial tree seen during bronchoscopy. Blood is visible at the entrance to the left upper lobe and lingual.

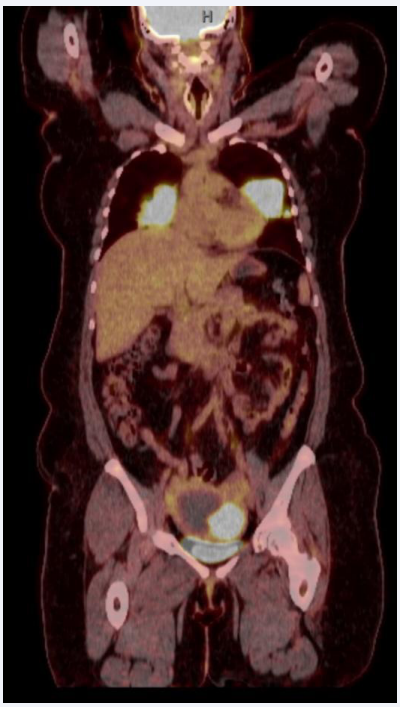

Figure 3 PET scan showing increased metabolic activity in lung nodules and uterine masses.

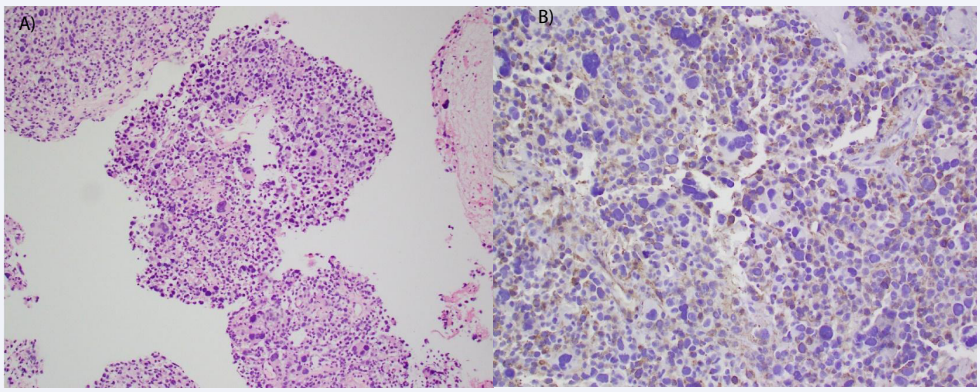

Figure 4 A) H&E stain of biopsy specimen obtained from a right pleural nodule showing scattered mitotic figures and significant cytologic atypia with large nuclei and prominent nucleoli B) immunohistochemical stain showing positive SMA staining in tumor cells.

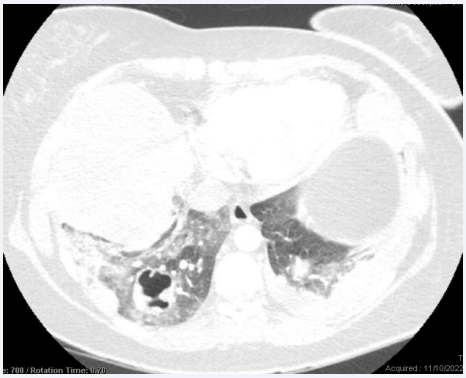

Figure 5 Repeat CT showing evolving lung masses with cavitation.

Society for Reproductive Medicine but reexamination typically occurs when patients experience change in their symptoms [8,9]. Leiomyosarcoma should be considered in postmenopausal women who develop rapidly enlarging fibroids, new onset pelvic pain, or bleeding in existing or new fibroids. Of note, neither of these features alone is a reliable predictor of leiomyosarcoma [2]. In addition to postmenopausal state, other risk factors for uterine cancer include nulliparity, late menopause, unopposed estrogen stimulation, feminizing ovarian tumors, tamoxifen exposure, obesity, diabetes mellitus, and family history of hereditary non-polyposis colorectal cancer [8]. Although the presence of malignant disease can be speculated based on these risk factors, a definitive diagnosis of uterine leiomyosarcoma cannot be made without histological examination, which requires surgical intervention and may conflict with the patient's desire to preserve fertility [2, 10].

To address this issue, our medical center conducted a study to create a risk prediction model for occult uterine sarcoma that could be used for women undergoing hysterectomies for presumed fibroids. The objective was to estimate risk of uterine sarcoma preoperatively. The clinical characteristics included in the final prediction model included age, race, body mass index (BMI), number of myoma, uterine weight, extent of fibroid growth, degree of pelvic pain, and need for blood transfusion within three months before hysterectomy. Although multiple features were strongly associated with uterine sarcoma, they failed to reliably predict disease when incorporated into this model. However, the authors suggest that the model may be useful in a subpopulation of patients who have higher baseline risk of uterine malignancy [3].

Perhaps the patient presented in this report falls into this subpopulation of individuals. She has multiple risk factors: nulliparity, obesity, and diabetes mellitus. She also meets criteria for some of the prediction model characteristics: uterine growth >50%, worsening pelvic pain requiring narcotics, and need for blood transfusions though she did not receive any because she is a Jehovah's Witness. Although the prediction model is not perfect, perhaps these findings could have increased clinical suspicion of uterine leiomyosarcoma, in which case the patient may have opted for a hysterectomy rather than uterine artery embolization and her disease could have been diagnosed before it spread to her lungs.

This patient's case and current existing literature highlight the difficulty of diagnosing uterine leiomyosarcomas. Our center's prediction model for uterine sarcoma is relatively novel and though it has its limitations, it may be useful for recommending more frequent surveillance of fibroids for those who develop clinical features included in the model. That is, if annual surveillance is recommended for patients with average risk, then patients with high-risk features as suggested by the prediction model may benefit from follow-up every three to six months. This is purely speculation at the present time, and it is unclear if this method of surveillance would allow for earlier detection of disease without causing harm. More research on imaging methods and other predictive markers would be helpful in advancing the management of this disease.t

ACKNOWLEDGEMENTS

The authors would like to thank our medical center's pathologist Dr. Aviv Hever for providing us with images of the patient's pathology slides.

REFERENCES

1. Roberts ME, Aynardi JT, Chu CS. Uterine leiomyosarcoma: A review of the literature and update on management options. Gynecol Oncol. 2018; 151: 562-572.
2. Vilos George A, Allaire C, Laberge PY, Leyland N. The Management of Uterine Leiomyomas. J Obst Gynaecol. 2015; 35: 157-178.
3. Lentz SE, Zaritsky E, Tucker LY, Lee C, Lazo IM, Niihara A, et al. Prediction of Occult Uterine Sarcoma before Hysterectomy for Women with Leiomyoma or Abnormal Bleeding. J Minim Invasive Gynecol. 2020; 27: 930-937.e1.
4. Multinu F, Casarin J, Tortorella L, Huang Y, Weaver A, Angioni S, et al. Incidence of sarcoma in patients undergoing hysterectomy for benign indications: a population-based study. Am J Obstet Gynecol. 2019; 220: 179.e1-179.e10.
5. Miettinen M. Immunohistochemistry of soft tissue tumours - review with emphasis on 10 markers. Histopathology. 2014; 64: 101-118.
6. "Uterine Sarcoma Survival Rates: American Cancer Society." Uterine Sarcoma Survival Rates | American Cancer Society.
7. Van den Bosch T, Coosemans A, Morina M, Timmerman D, Amant F. Screening for uterine tumours. Best Pract Res Clin Obstet Gynaecol. 2012; 26: 257-266.
8. Florence AM, Fatehi M. Leiomyoma. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022.
9. Parker WH. Uterine myomas: management. Fertil Steril. 2007; 88: 255-271.
10. Arleo EK, Schwartz PE, Hui P, McCarthy S. Review of Leiomyoma Variants. AJR Am J Roentgenol. 2015; 205: 912-921.