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# Recurrent benign leiomyomas after total abdominal hysterectomy. Rich or poor estrogenic environment may lead to their recurrence?

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## ABSTRACT

**INTRODUCTION:** Benign metastasizing leiomyomas represent benign lesions consisted by leiomatous tissue and could be observed in positions away from their usual localization, the human uterus. They commonly affect women that have undergone total hysterectomy. Approximately 100 similar cases have been reported in the literature, so the case we present is rare and reviewing the literature and needs to be reported.

**PRESENTATION OF CASE:** We report a case of a 55 year old Greek woman, gravida five and para three, who attended our unit 3 years ago complaining of occasionally lower abdominal pain and irritation the last months. Fourteen years ago she underwent abdominal hysterectomy and left salpingo-oophorectomy due to a 13 cm uterine leiomyoma. In the meantime she underwent two surgical procedures for recurrent benign leiomyomas.

**DISCUSSION:** When patient was admitted at this time, clinical examination revealed a palpable mass of 5 cm. The transvaginal ultrasonography revealed 3 masses in the lower pelvis of unknown origin. The patient underwent a new laparotomy revealing three masses of benign leiomyomas with low mitotic activity.

**CONCLUSION:** Our case supports the recurrent appearance of leiomyomas in pelvis after total abdominal hysterectomy and is one of few reports in literature where the tumors appear in the same patient both in estrogen rich and estrogen poor environment. Additionally, we show the importance of transvaginal ultrasonography and 3 dimensional power Doppler in the differential diagnosis of pelvic masses. Thus, transvaginal ultrasonography seems to be a pivotal tool for the diagnosis and follow up of these challenging lesions.

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## 1. Introduction

Benign metastasizing leiomyomas represent benign lesions consisted by leiomatous tissue; the main difference from uterine leiomyomas is the fact that they could be observed in positions away from their usual localization, the human uterus.

The term BML was firstly described in 1939 by Steiner through a case of a 36 yrs old woman who died due to the lethal pulmonary

metastasis of a solid mass. This solid feature was shown to be consisted by leiomyomatous tissue.

Histologically these benign tumors consist of smooth muscle fibers separated with fibers consisting of connective tissue [1] and as reported they can behave as malignant and metastasize in various areas such as pelvis, lungs, retroperitoneally and even in inferior vena cava, right atrium and limbs. The most common site of metastases is the lung [2]. This rare entity commonly affects women that have undergone total hysterectomy and can cause symptoms that range from mild to severe depending on the size of the tumor. Approximately 100 cases have been reported in the literature.

We report a case of a female patient with benign metastasizing leiomyomas (BML). The patient underwent total abdominal hysterectomy and left salpingo-oophorectomy for benign leiomyomas and in the following years 3 additional laparotomies for relapsing BML. This work has been reported according to the SCARE criteria [3].

**Abbreviations:** BML, benign metastasizing leiomyoma; 3D, 3 dimensional; IVP, intravenous pyelogram; RBC, red blood cells; FFP, fresh frozen plasma; MRI, magnetic resonance imaging; TVS, transvaginal ultrasound; TAH, total abdominal hysterectomy; HPF, high power field; GnRH, gonadotropin –releasing hormone.

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Fig. 1. 3D ultrasonographic appearance of the two masses.



Fig. 2. 3D ultrasonographic appearance of the third mass.

## 2. Case presentation

A 55-year-old Greek female patient, gravida five and para three, was admitted to our clinic 3 years ago with symptoms of occasionally lower abdominal pain and irritation since some months ago. Her obstetric history restrained to 3 normal deliveries and 2 pregnancy terminations due to social-economic reasons. Her last menstrual period was at the age of 44, her Pap smear tests and her mammography were negative. Her mother was diagnosed with breast cancer at the age of 60. Her personal medical history was significant for hypertension, diabetes type I and mild rise in cholesterol the last months, while her psychosocial history was uneventful. She was non smoker with no drug using. She was 1,65 m and 88 kgr in weight (BMI 32,3). The patient had an interesting history of successive abdominal operations. Fourteen years ago she underwent open abdominal hysterectomy and left salpingo-oophorectomy due to a 13 cm uterine leiomyomas without morcellating it. After almost 4 years from the first operation, she underwent surgery for an abdominal mass close to the remaining right ovary and another mass with vaginal cuff origin. The right ovary was also removed in this laparotomy. Histological diagnosis revealed three leiomyomas with low mitotic activity (0–1/10 HPF). Three years after the last surgery, a recurrence of a tumor originating from the vaginal cuff was presented. A new surgical procedure was performed and a leiomyoma was confirmed via histological results.

When the patient was admitted at this time, a clinical examination was performed, revealing a palpable solid, painful and immobile mass of approximately 5 cm. This mass was located at the site of the right uterosacral ligament in the transrectal examination. The female patient was then screened via transvaginal ultrasound revealing 3 masses in the lower pelvis of unknown origin (Figs. 1 and 2).

The tumors had excessive vascularization via application of 3D power Doppler. The neo-vessels were presented with multiple branches of blind ends.

The use of 3D power Doppler has improved the assessment of the vascularity in case of a tumor. Nevertheless this technique has not been studied thoroughly as far as fibroids are concerned [4].

The IVP of the urinary system and the chest x ray were both normal. The laboratory tests were normal. Finally the patient underwent a new laparotomy that revealed three masses by an experienced gynecologist in gynecological oncology. The origin of the first mass (6 cm) was the vaginal cuff. The second mass of 5 cm was located in the right pararectal area and the third mass of 7 cm was located in the left pararectal area and was removed en block with part of rectosigmoid colon due to solid adhesions. An end to end intestinal anastomosis was then performed. The total duration of the operation was about 6 h and the patient received 4 RBC units, 2 FFP and approximately 10 lt of i.v fluids. There were no peri-operative and postoperative complications and the patient had a good recovery; she was followed up in a simple ward and she was dismissed during the following days. The patient was compliant with the therapeutic program which was well tolerated, with no significant side-effects or hospitalizations. The histology showed that all three masses were also benign leiomyomas with low mitotic activity. The patient is in excellent clinical condition and she is followed up via transvaginal ultrasonography every year without any finding after the last surgery.

## 3. Discussion

Leiomyomas are benign, monoclonal, mesenchymal tumors that usually originate from uterine smooth muscle cells. They can appear as distant metastasis in anatomic areas such as lungs, vessel structures even retroperitoneally. It is reported that simple leiomyomas reappear at a rate of 5,7–62% at 5 years after myomectomies [1]. BML is a rare entity that usually appears in women that have undergone hysterectomy for leiomyomas [2].

Despite that leiomyomas appear often in general population, metastatic leiomyomas are very rare and appear usually after total hysterectomy and bilateral salpingo-oophorectomy. Yarci et al. presented an interesting case of recurrent leiomyomas after total hysterectomy and bilateral salpingo-oophorectomy without hormone replacing therapy with estrogen [5]. In our case the leiomyoma appeared for the first time 4 years after the initial hysterectomy and grew in a estrogenic environment as the right ovary was preserved. The consequent leiomyomas grew in a estrogen poor environment because the right ovary was removed through the second laparotomy and no hormone replacement therapy was received.

The symptoms of BML are restrained only to compression of the nearby organs whereas leiomyomatosis of the lungs is usually accidentally identified in routine chest X-ray [6]. There is strong indication that BML is hormone-dependent. The presence of estrogen and progesterone receptors has been clearly documented [7]. According to these observations it is reasonably assumed that tumor growth could be controlled with bilateral oophorectomy [8]. Other reasonable method would be the use of long-acting GnRH analogs who would suppress the endogenous gonadotropin secretion which is necessary for gonadal steroid production [9]. Progesterone has also been reported to be effective in cases of leiomyomatous tumors [10].

Diagnosis of leiomyomas can sometimes be challenging due to the fact that they can extend away from the uterus, where their typical area of origin is, and mimic a malignant tumor. Methods that are usually used are vaginal ultrasonography, MRI and hysteroscopy. A more recent study reports a sensitivity of TVS for the diagnosis of leiomyoma of 96.38% and specificity 96.00% [11]. The typical appearance of the uterine leiomyoma is a solid tumor with whorled appearance and the same echogenicity of the surround-

ing myometrium even though sometimes it can appear hypoechoic. Usually acoustic shadows may be presented making the leiomyoma appearing like dispersing in the surrounding tissues. Via Doppler software application, leiomyoma appears to have a circumferential vascularity and high flow rates with Doppler but in cases of necrosis or degeneration this type of vascularity is usually absent [12]. This high velocity flow in blood vessels along with low resistance index is a major characteristic in newly formed vessels in cases of neovascularization [13]. The diagnosis of leiomyoma is highly dependent upon the sonographer's experience. For this reason in cases of doubt, appropriate referral of the patient should be made.

In conclusion, BML is a rare entity that usually affects women after hysterectomy for leiomyomas. Our case supports the recurrent appearance of leiomyomas in pelvis after TAH and is one of few reports in literature where the tumors appeared in the same patient both in estrogen rich and estrogen poor environment. It also adds new entity to the differential diagnosis of adnexal masses.

### Conflicts of interest

NA.

### Funding

NA.

### Ethical approval

IRB/Ethics Committee ruled that approval was not required for this study.

### Consent

Written informed consent was obtained from the patient, and this has been stated in the manuscript per the submission instructions.

### Author contribution

Conception and design the study: Konstantinos Kyriakopoulos, Sofoklis Stavrou.

Acquisition of data: Konstantinos Kyriakopoulos, Ekaterini Domali, Sofoklis Stavrou.

Analysis and/or interpretation of data: Ekaterini Domali, Konstantinos Kyriakopoulos.

Drafting the manuscript: Konstantinos Kyriakopoulos, Ekaterini Domali, Sofoklis Stavrou, Alexandros Rodolakis, Peter Drakakis.

Revising the manuscript critically for important intellectual content: Peter Drakakis.

Approval of the version of the manuscript to be published: Konstantinos Kyriakopoulos, Ekaterini Domali, Sofoklis Stavrou, Alexandros Rodolakis, Peter Drakakis.

### Registration of research studies

As this is not a 'first-in-man' case study, our paper is not eligible to be registered.

### Guarantor

Konstantinos Kyriakopoulos.  
Ekaterini Domali.  
Sofoklis Stavrou.  
Alexandros Rodolakis.  
Peter Drakakis.

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