

# Retroperitoneal Leiomyoma Metastases: A Rare Entity In The Differential Diagnosis Of Adnexal Masses

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## Abstract

Histologically benign uterine smooth muscle tumors can behave as malignant and can spread usually to the lungs and rarely to retroperitoneal structures, peritoneum, lymph nodes, bone and soft tissues. Women who have undergone hysterectomy for leiomyomas are most commonly affected. We report a case of bilateral retroperitoneal leiomyoma metastases 12 years after hysterectomy. Bilateral retroperitoneal leiomyoma metastasis was not reported before. This case report also adds a new entity to the differential diagnosis of adnexal masses.

## INTRODUCTION

Histologically benign uterine smooth muscle tumors are reported to behave as malignant and spread to the lungs, pelvic, retroperitoneal lymph nodes, omentum, inferior vena cava, right atrium and limbs (1).

Metastasizing leiomyoma is a rare entity that most commonly affects women after hysterectomy and is usually asymptomatic at presentation. The average interval between surgery for the initial uterine leiomyoma and detection of distant metastasis is reported to be 10 years (1).

Estrogen was claimed to cause growth of metastasizing leiomyomas and prognosis is worse in the premenopausal women. Identification of estrogen and progesterone receptors in the lung lesions have led to development of therapeutic options based on hormonal manipulation with surgical or medical oophorectomy (2).

We report a case operated for bilateral adnexal masses 12 years after hysterectomy and found to have bilateral retroperitoneal leiomyoma metastases.

## CASE REPORT

A 53-year-old woman was admitted to our hospital with the complaint of hot flushes. Bilateral adnexal masses were found at routine gynecologic examination. Her medical history revealed a myomectomy operation 25 years ago for uterine leiomyoma of 6 cm in size. By that time she already had two normal spontaneous labors. Two years after this surgery, the patient gave birth to her third child with

cesarean section. 13 years ago, the patient presented with menorrhagia and total abdominal hysterectomy was done with the diagnosis of recurrent uterine leiomyoma. Pathologic examination of the specimen revealed one intramural and one submucous uterine leiomyoma, 6 cm and 4 cm in size respectively. After this operation, the patient had no gynecologic examination.

Bilateral adnexal fullness was detected at gynecologic examination. Transvaginal ultrasonography confirmed bilateral adnexal solid masses, 64 x 86 mm on the right and 56 x 53 mm on the left. All routine laboratory tests were normal including AFP: 7ng/ml,  $\beta$ -HCG: 3.6mUI/ml, Ca 199: 9U/ml and Ca 125: 25U/ml. These findings led us to plan laparotomy with the diagnosis of bilateral adnexal masses.

Lower abdominal midline incision was preferred in case of malignancy. On exploration of the pelvis, 8cm mass was detected on the right retroperitoneal region, superior to the psoas muscle and at the back of the infundibulopelvic ligament, on iliac bifurcation. On the left side, 5cm retroperitoneal mass was below the infundibulopelvic ligament, reaching from external iliac vein to the obturator fossa. Peritoneum over the mass at right side was opened and right ureter was observed. The mass was extracted with sharp and blunt dissections and sent for frozen section. Frozen report was leiomyoma. After getting this result, right ovary and fallopian tube were observed and right salpingoophorectomy was done. On the left side, the peritoneum over the mass was opened. After dissection of left ureter, infundibulopelvic ligament and external iliac vein

and freeing the mass from surrounding tissues with sharp and blunt dissections, the mass was extracted. Surgery was completed following left salpingoophorectomy.

Pathologic examination has revealed bilateral leiomyomas showing degenerative changes. Tumor had 0-2 mitoses / 50 high power field (HPF). There was no nuclear atypia or coagulative necrosis. Both ovaries and fallopian tubes were normal.

### **DISCUSSION**

Metastasizing benign leiomyoma is a rare entity which is usually seen in women who have undergone hysterectomy for leiomyomas (1). Retroperitoneal leiomyomas probably originate from hormone sensitive smooth muscle cells. They may be metastases of uterine leiomyomas or primary soft tissue tumors. They are distinguished from parasitic leiomyomas because they are distant from the uterus (2).

The pathogenesis of benign metastasizing leiomyoma has been a subject of controversy over the years. In the past, metastatic foci in the lungs were thought to originate in situ in the lungs and were named as "multiple fibroleiomyomatous hamartomas" (3). Today, most pathologists accept these lesions as hematogenous metastases from benign uterine leiomyomas, but controversy still exists for the lesions with increased mitotic activity.

Retroperitoneal leiomyomas can be differentiated from leiomyosarcomas histologically with their low mitotic activity and absence of nuclear atypia and necrosis (3). Additionally, retroperitoneal leiomyomas are usually positive for estrogen and progesterone receptors (3). Most of these tumors seem to have good long-term prognosis with a small potential for local recurrence. Although some authors have proposed metastasizing benign leiomyomas as low-grade leiomyosarcomas (4), they are usually accepted as benign tumors because of their pathologic features, prognosis and response to hormonal therapy. Our case also shows benign pathologic features with low mitotic activity. The argument that more extensive sampling of uterine tissue may reveal sarcomatous degeneration, does not explain the benign histology of metastatic foci.

Although uterine leiomyomas are very frequent in the general population, metastasizing leiomyoma is very rare. Several theories have been improved for the mechanism of spread. One of these is the spread of the smooth muscle cells after uterine extension into the pelvic venous channels (5). It has been also proposed that smooth muscle tumors may arise

directly from vessel walls. However this is associated with extension into the inferior vena cava and does not result in metastatic lesions. This entity is termed as "intravenous leiomyomatosis" (6). Others propose that tumor gain venous access from surgical trauma during hysterectomy (6). But this does not explain cases presented with metastatic foci before hysterectomy.

In the differential diagnosis of metastasizing leiomyoma to retroperitoneal soft tissues; leiomyoma metastases to retroperitoneal lymph nodes, leiomyoma of the urinary bladder, intravenous leiomyomatosis and leiomyomatosis peritonealis disseminata should be remembered.

Intraoperative exploration and histologic examination are crucial for the definite diagnosis. We were not able to find another case report in the literature telling about bilateral retroperitoneal leiomyoma metastases. Bilateral cases may be confused with ovarian tumors as in our case, or leiomyoma of the broad ligament.

Retroperitoneal leiomyomas may be completely asymptomatic or may cause a range of symptoms from abdominal pain to hydronephrosis due to urethral obstruction. For both diagnosis and treatment, first of all surgical extraction of the tumor is needed. For appropriate cases, total abdominal hysterectomy and bilateral salpingoophorectomy can be added at the time of operation. Hormonal therapy for cases without oophorectomy, to prevent local recurrence is controversial. It is reported that hormonal therapy with progesterone, tamoxifen and gonadotropins to remove estrogen source is associated with tumor regression (7). In our case, there was no need to concern hormonal therapy as bilateral oophorectomy was done.

### **CONCLUSIONS**

In conclusion, benign metastasizing leiomyoma is a rare entity that usually affects women after hysterectomy for leiomyomas and it is seen most frequently in the lungs. Our case supports the presence of retroperitoneal leiomyoma metastasis from the uterus and is the first reporting of bilateral involvement. It also adds a new entity to the differential diagnosis of adnexal masses.

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