

Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation

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ABSTRACT

Uterine leiomyomas (fibroids) are common benign neoplasms, which develop from the muscular tissue of the uterus with an estimated incidence of 20-40% in women of reproductive age. In the early nineties, power morcellators were introduced and became commonly used during hysterectomy for symptomatic fibroids. However, if all fragments are not removed, they may parasitize to other blood supply and present as abdominal or pelvic masses. Unfortunate cases have also been reported in which uterine sarcomas seeded throughout the abdomen and pelvis secondary to morcellation. The Food and Drug Administration (FDA) estimates that 1 in 350 women undergoing hysterectomy or myomectomy for fibroids is found to have an unsuspected uterine sarcoma. As a result, the FDA issued a press release in 2014 discouraging the use of power morcellators. Recently, the FDA approved a new containment device, the PneumoLiner, for use with certain power morcellation devices. However, it is unknown if this device will help to reduce the risk of seeding fibroids and unsuspected uterine malignancies. We present a case in which a patient who underwent morcellation therapy for symptomatic fibroids presented with recurrent abdominal and pelvic leiomyomas mimicking malignancy.

CASE REPORT

CASE REPORT

A 50-year-old female with a past medical history of uterine fibroids status post robotic hysterectomy with morcellation presented with acute abdominal pain. She had a noteworthy family history of uterine leiomyosarcoma and lymphoma.

The patient was initially diagnosed with multiple uterine fibroids via ultrasound in December 2010 [Figure 1], the largest of which was posteriorly located and measured 6.7 x 5.8 x 4.9 cm. Conservative management and routine follow up was planned. The patient returned for follow up ultrasound in June 2011 [Figure 2], which demonstrated interval enlargement of the fibroids now measuring up to 8.3 cm in

greatest dimension. A robotic approach was not being offered at that time at this institution. Therefore, she sought evaluation at an outside institution where she ultimately underwent a robotic hysterectomy with a power morcellator.

Subsequently, she presented years later in November 2015 with acute onset of pelvic pain. A transvaginal ultrasound demonstrated a solid vascular mass within the pelvis measuring 10.2 x 5.9 cm [Figure 3]. Given the patient's history of robotic hysterectomy with morcellation and her family history of uterine leiomyosarcoma, the possibility of seeding from an undiagnosed leiomyosarcoma was raised. The patient subsequently underwent a CT scan of the abdomen and pelvis in December 2015 [Figures 4a-c], which demonstrated a large, lobulated, solid soft tissue mass in the pelvis as well as

suspected bilateral iliac adenopathy and several smaller abdominal masses. Based on these findings and her family history, lymphoma was added to the differential diagnosis.

The patient was then referred for CT guided core biopsy of the dominant pelvic mass [Figure 5]. Pathology revealed a cytologically bland smooth muscle neoplasm with focal epithelioid features and no mitotic figures or necrosis. Flow cytometry revealed no evidence of lymphoma. The patient subsequently underwent laparotomy with removal of eleven masses, all of which revealed similar pathology to the percutaneous core biopsies and was consistent with multiple intraperitoneal leiomyomas. A follow up MRI of the abdomen and pelvis approximately 8 months later revealed no evidence of recurrent or residual masses.

DISCUSSION

Etiology & Demographics:

Uterine leiomyomas are common benign uterine neoplasms, which develop from the muscular tissue of the uterus in approximately 20-40% of women of reproductive age. These growths are estrogen dependent and therefore will often spontaneously regress in post-menopausal women. Early menarche, obesity, alcohol use, multiparity, and ovulation induction agents all pose risk factors for the formation of uterine fibroids. The prevalence of these tumors has been estimated to be as high as 77% [1, 2]. Uterine fibroids are among the most common indications for both pelvis ultrasound and hysterectomy in the United States. Disseminated Peritoneal Leiomyomatosis is a rare disease entity in which multiple smooth muscle implants are noted through the peritoneum of the abdomen and pelvis. This is a known complication in patients who have undergone hysterectomy with power morcellation [1]. Fibroids may either be intramural, subserosal, submucosal, or pedunculated. Submucosal fibroids are known to cause excessive menstrual bleeding, which can cause prolonged menstrual cycles. In contrast, subserosal fibroids do not tend to cause abnormal or excessive menstrual cycles.

The FDA estimates that 1 in 350 women undergoing hysterectomy or myomectomy for fibroid treatment is found to have an unknown uterine sarcoma [3, 4, 5]. Therefore, laparoscopic power morcellation poses a risk of spreading unsuspected cancerous and noncancerous tissue beyond the uterus into the pelvic and abdominal cavities. As a result, the Food and Drug Administration issued a press release in April 2014 that discouraged the use of power morcellators. Recently, the FDA approved a new containment device, the PneumoLiner, for use with certain power morcellation devices. However, it is unknown if this device will help to reduce the risk of seeding fibroids as well as unknown uterine cancers. Continued research is warranted to assess this risk.

Clinical & Imaging findings:

In most cases, these growths are asymptomatic. However, in some women they are symptomatic and can cause heavy or prolonged vaginal bleeding, pelvic pain, and/or bulk related

symptoms such as frequent urination. As in the case presented, acute abdominal pain is often the presenting symptom.

We present the case of a 50-year-old woman who underwent robotic hysterectomy with power morcellation and was later found to have multiple benign fibroids mimicking malignancy throughout her abdomen and pelvis. Recognition of this entity and associated imaging findings is an important key for proper diagnosis. Tissue diagnosis may be required to exclude malignancy in these cases. Uterine fibroids may be evident on plain film, when calcified, however, ultrasound is often first line. On ultrasound, fibroids appear as a homogenous, hypoechoic mass with or without shadowing. The uterus may be enlarged and lobulated. Color Doppler will show marked peripheral flow with decreased central flow.

Treatment & Prognosis:

Many treatment options are available for symptomatic uterine fibroids including medical, endovascular, or surgical therapy. Among the surgical options, laparoscopic hysterectomy with power morcellation became popular in the late nineties owing to its shorter post-operative recovery time, ability to perform less invasive surgery and the potential reduced risk of complications compared to open abdominal hysterectomy and myomectomy.

A laparoscopic power morcellator is a medical device with a fast rotating cylindrical knife that divides tissue into smaller pieces or fragments in order to assist removal of the tissue through small laparoscopic incision sites [6]. Careful attention must be made to remove all of the uterine fragments. If all fragments are not removed, they may parasitize to other blood supply and present as abdominal or pelvic masses [3, 7]. Other unfortunate cases have also been reported in which a uterine sarcoma seeded throughout the abdomen and pelvis secondary to morcellation.

Differential Diagnosis:

The main differential considerations for uterine masses include but are not limited to uterine leiomyoma, ovarian fibroma, uterine adenomyoma, uterine leiomyosarcoma, and endometrial carcinoma. Specifically, when it comes to power morcellation, uterine leiomyoma and uterine leiomyosarcoma seeding have been reported in the literature.

Uterine Leiomyoma

Uterine leiomyomas are the most common uterine mass. While generally benign, small percentages have been reported to undergo malignant degeneration. Approximately 20% to 80% of women develop fibroids by the age of 50. Hysterosalpingography may demonstrate mass effect on the adjacent endometrium, especially if they are submucosal in origin. Uterine leiomyomas are most commonly diagnosed on ultrasound, where they appear as homogenous hypoechoic masses with or without shadowing. On CT, they appear as homogeneously isodense masses within the uterus, which often cause a contour deformity. Dystrophic calcifications may be present. They appear as isointense to myometrium on T1WI and hypointense to myometrium with a hyperintense rim on T2WI. FDG PET/CT does not show increased radiotracer uptake.

Uterine Adenomyoma

An adenomyoma is a focal region of adenomyosis (ectopic endometrial tissue within the myometrium), which results in a mass, which is often indistinguishable from uterine leiomyoma. Adenomyosis often affects women of reproductive age and is seen more commonly in women who have undergone uterine procedures such as cesarean section or dilation & curettage. Adenomyoma implants are generally not a risk for patients who have undergone hysterectomy with power morcellation. Hysterosalpingography may show mass effect on the subjacent endometrium. On ultrasound, focal adenomyosis appears as a hypoechoic and heterogeneous myometrial mass with ill-defined borders. On MRI, they are isointense to myometrium on T1WI with occasional high signal due to foci of small areas of hemorrhage. On T2WI, they appear as circumscribed, ill-defined, low signal intensity myometrial masses. FDG PET/CT does not show increased radiotracer uptake.

Ovarian Fibroma

An ovarian fibroma is a benign sex cord-stromal tumor of the ovary, which makes up approximately 4% of ovarian neoplasms. In a patient with a pelvic mass status post hysterectomy with morcellation, an ovarian fibroma should be considered. Ovarian fibromas appear hypoechoic on ultrasound and isodense to the uterus with or without calcifications on CT, much like uterine leiomyoma. On T1WI and T2WI, they are homogenous and isointense-to-hypointense to the uterine myometrium.

Uterine Leiomyosarcoma

Uterine leiomyosarcoma is a rare aggressive cancer of the uterus, accounting for approximately 8% of all uterine malignancies. Patients who have undergone hysterectomy with power morcellation for uterine leiomyosarcoma have been reported to have cancerous seeding of leiomyosarcoma throughout the peritoneum. This entity should always be considered in a patient status post hysterectomy with power morcellation who presents with peritoneal or pelvic masses. They appear as heterogeneous, ill-defined masses on ultrasound, and will often demonstrate increased vascularity on power Doppler imaging. On CT, they are heterogeneously enhancing, low attenuating (relative to the homogeneously enhancing myometrium) masses. On T1WI, they demonstrate low to intermediate signal intensity with areas of hemorrhage, which appear as high T1 signal intensity. On T2WI, they are intermediate signal intensity with areas of high signal intensity due to necrosis. These cancers have avid FDG uptake in primary and metastatic lesions.

TEACHING POINT

One option for hysterectomy due to symptomatic fibroids includes laparoscopic use of power morcellation. If all fragments are not removed, they may parasitize to other blood supplies and present as abdominal or pelvic masses. Intraperitoneal leiomyomas should be included in the differential diagnosis of multiple abdominal and pelvic masses in patients with a history of prior morcellator-assisted hysterectomy.

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FIGURES

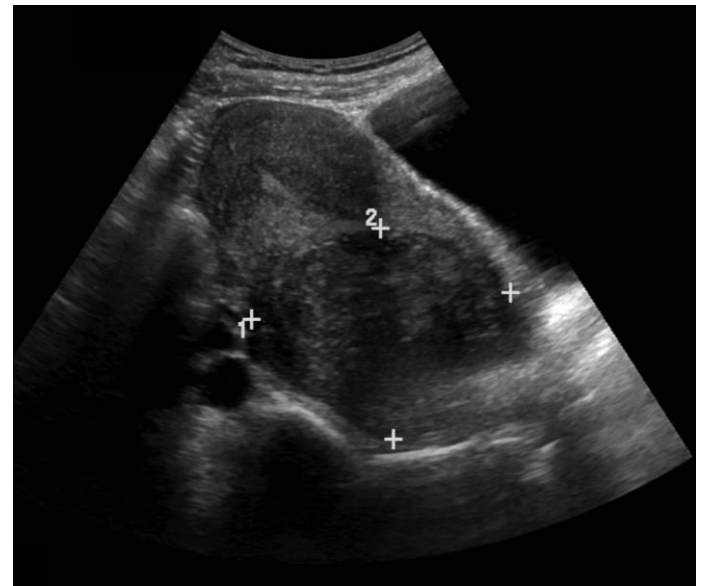
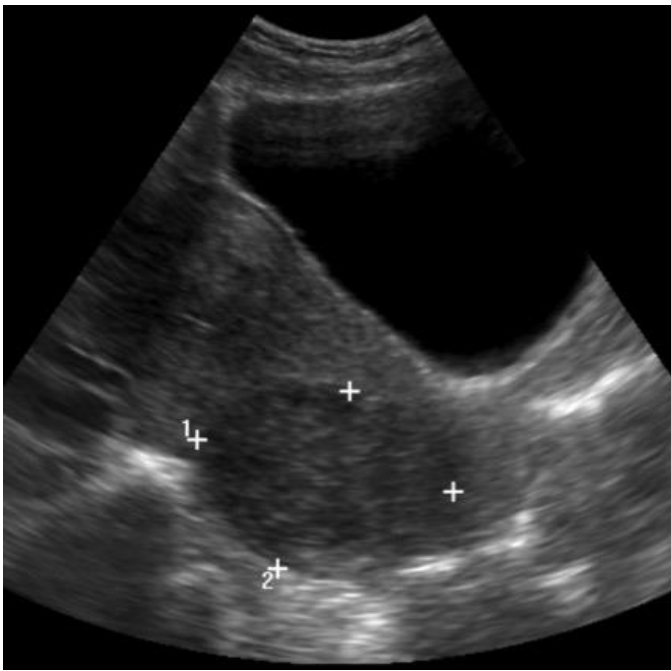


Figure 2: 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation. Findings: Transabdominal ultrasound of the uterus in the sagittal plane same patient six months later, demonstrates the same posteriorly located fibroid [cursors] now measuring up to 8.2 cm in greatest dimension. Technique: B-mode Transabdominal sonogram of the pelvis. Sagittal section of the uterus. Ultrasonography was performed using 3-5MHz curvilinear transducer.

Figure 1: 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation. Findings: Transabdominal ultrasound of the uterus in the sagittal plane at original presentation for pelvic pain, demonstrates a posteriorly located fibroid [cursors] measuring up to 6.7 cm. Technique: B-mode Transabdominal sonogram of the pelvis. Sagittal section of the uterus. Ultrasonography was performed using 3-5MHz curvilinear transducer.

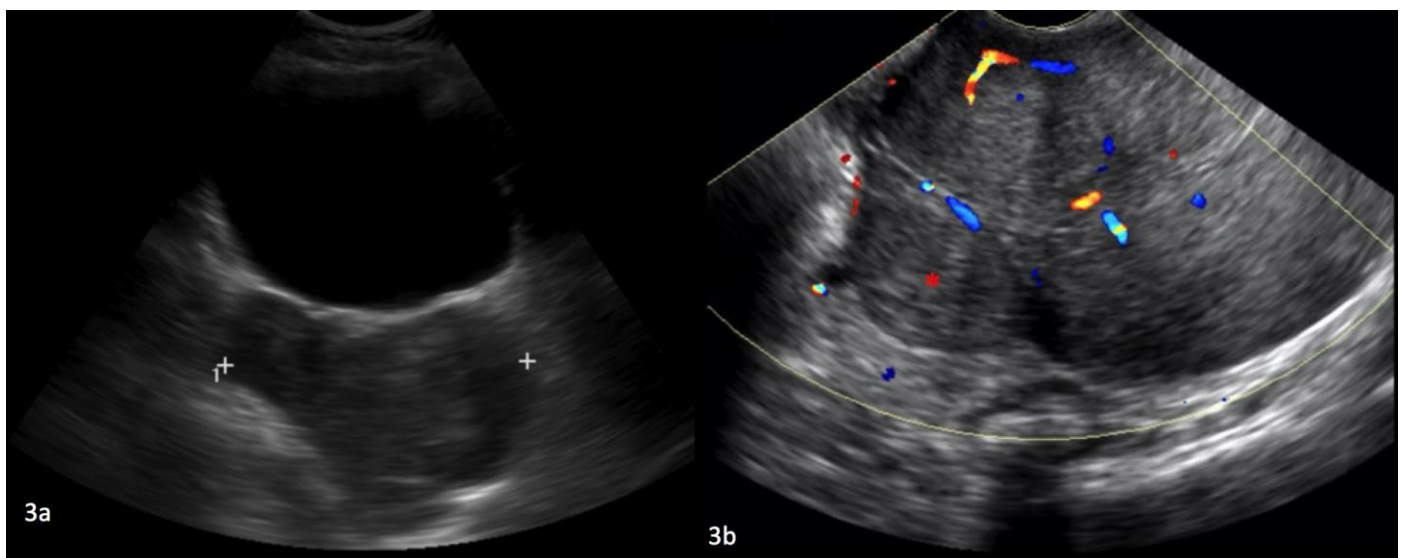


Figure 3: 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation. Findings: a) Transabdominal ultrasound image of the pelvis in the same patient several years after robotic hysterectomy with power morcellator demonstrates a 10.3 cm lobulated, solid mass [cursors] within the pelvis. Technique: B-mode Transabdominal sonogram of the pelvis. Ultrasonography was performed using 8MHz curvilinear transducer. b) Transvaginal ultrasound image of the pelvis in the same patient several years after robotic hysterectomy with power morcellator. A lobulated, solid, vascular mass is identified within the pelvis [asterisk]. Technique: Power Doppler Transvaginal sonogram of the pelvis using a 3-5MHz IVUS transducer.

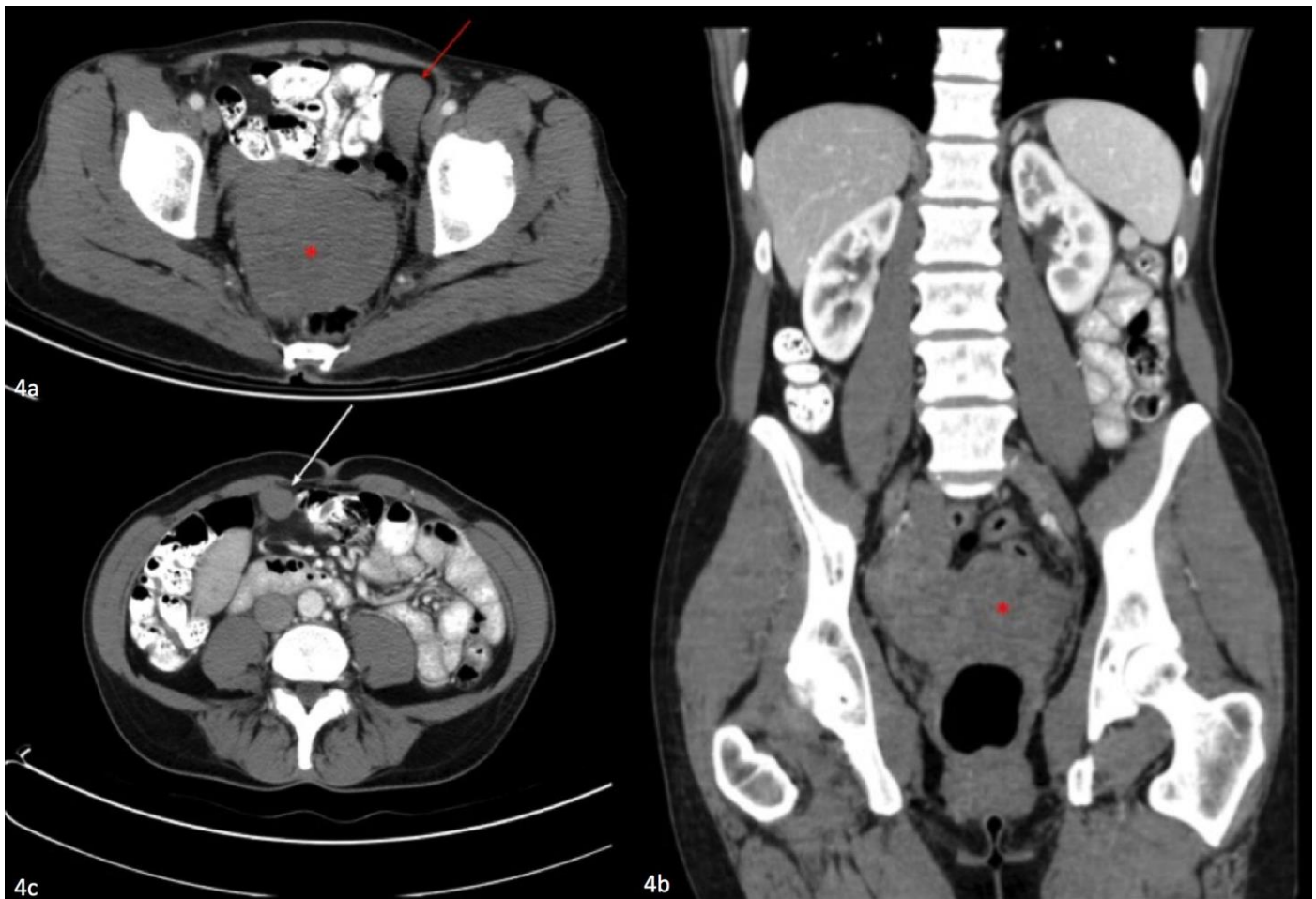


Figure 4: 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation.

Findings:

a) Axial contrast enhanced CT of the pelvis demonstrates the solid nonenhancing, homogenous pelvic mass [asterisk], with well-defined margins, as well as multiple other masses including a left external iliac mass [red arrow].

Technique: mA 250, kvp 130, 5.0 mm slice thickness, with 100ml, Omnipaque 350.

b) Coronal contrast enhanced CT of the abdomen and pelvis demonstrates the solid nonenhancing, homogenous pelvic mass [asterisk], with well-defined margins.

Technique: mA 250, kvp 130, 5.0 mm slice thickness, with 100ml, Omnipaque 350.

c) Axial contrast enhanced CT of the pelvis demonstrates an anterior intraperitoneal nonenhancing, homogenous abdominal mass [white arrow], with well-defined margins.

Technique: mA 250, kvp 120, 5.0 mm slice thickness, with 100ml, Omnipaque 350.

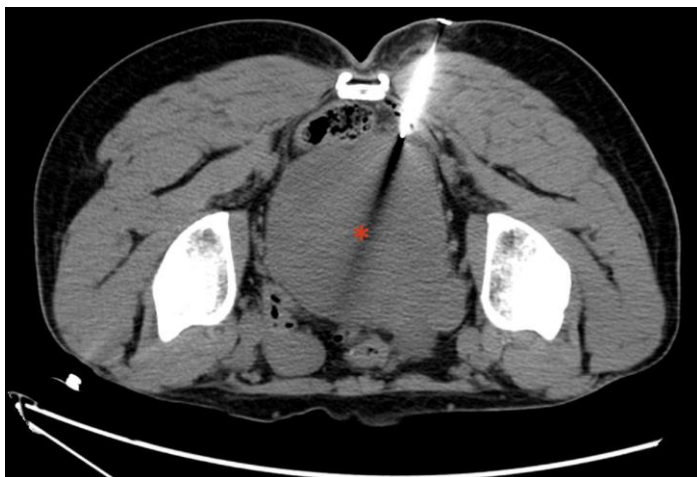


Figure 5 (left): 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation.

Findings: Noncontrast axial CT of the pelvis in the prone position demonstrates a CT-guided core biopsy of the large, homogenous pelvic mass [asterisk], with well-defined borders.

Technique: mA 360, kvp 120, 5.0 mm slice thickness.



Figure 6 (left): 50 year old female patient: Diagnosis of Disseminated Peritoneal Leiomyomatosis Status Post Laparoscopic Hysterectomy with Morcellation. Findings: Intra-operative picture demonstrating the largest leiomyoma [asterisk] attached to the transverse colon [red arrow].

Etiology	Unclear, likely multifactorial.
Incidence	Cumulative incidence of any size by age 50 is >80% for black women and >70% for white women [2].
Gender Ratio	Female predominant
Age Predilection	Reproductive age (black women - <30 years of age, white women - >30) [2].
Risk Factors	Early menarche, parity, ovulation induction agents, obesity, alcohol use [2].
Treatment	Surgery mainstay therapy, hysterectomy as definite procedure, with myomectomy performed through various techniques. Symptomatic relief prior achieved by combined hormonal contraceptives [1].
Prognosis	Myomectomy – up to 50% chance of recurrence, Hysterectomy – up to 12% of patients report new symptoms [1].
Image Findings	Transvaginal US: Homogenous hypoechoic mass with or without shadowing, enlarged lobulated uterus with color Doppler displaying marked peripheral flow with decreased central flow [3].

Table 1: Summary table for Uterine Leiomyoma.

Differential Diagnosis	X-Ray	US	CT	MRI	PET
Uterine Leiomyoma	Hysterosalpingography – mass effect on endometrium	Homogenous hypoechoic mass with or without shadowing enlarged lobulated uterus.	Homogenously isodense to myometrium with uterine contour deformity.	<u>T1WI</u> – isointense to myometrium, lobulated contour. <u>T2WI</u> – homogenous, well defined, hypointense to myometrium with hyperintense rim.	FDG PET/CT does not show increased radiotracer uptake.
Uterine Adenomyoma	Hysterosalpingography – mass effect on endometrium	Hypoechoic, heterogeneous myometrial mass with ill-defined borders.	Variable, nonspecific. “Swiss cheese” appearance due to lack of enhancement of dilated cystic glands.	<u>T1WI</u> – Isointense to myometrium, occasional high signal intensity foci due to small areas of hemorrhage. <u>T2WI</u> – Circumscribed, ill-defined, low signal intensity myometrial mass.	FDG PET/CT does not show increased radiotracer uptake.
Ovarian Fibroma	N/A	Hypoechoic mass with sound attenuation and edge shadows.	Nonspecific adnexal mass isodense to uterus with or without calcification.	<u>T1WI</u> – Homogenous, isointense to hypointense compared to uterine myometrium <u>T2WI</u> – same as T1WI	FDG PET/CT does not show increased radiotracer uptake.
Uterine Leiomyosarcoma	Hysterosalpingography – mass effect on endometrium	Heterogeneous echogenicity, with color Doppler showing increased vascularity.	Heterogeneously enhancing, low attenuation mass, relative to homogenous enhancing myometrium.	<u>T1WI</u> – Low to intermediate signal intensity mass, areas of hemorrhage show high T1 signal intensity. <u>T2WI</u> – Intermediate signal intensity heterogeneous mass. Necrosis shows high signal intensity.	Avid FDG-18 uptake in primary tumor and metastatic lesions.
Endometrial Carcinoma	Hysterosalpingography – mass within the endometrial cavity	Focal, thickened endometrium, areas of mixed echogenicity.	Endometrial thickening, hypoenhancing to myometrium.	<u>T1WI</u> – Hypo or isointense to normal endometrium/myometrium. <u>T2WI</u> – same as T1WI	FDG-18 useful for metastasis and surveillance.

Table 2: Differential Diagnosis table for Uterine Leiomyoma.

ABBREVIATIONS

cm: Centimeters
 CT: Computed Tomography
 FDA: Food & Drug Administration
 FDG-18: Fluorodeoxyglucose-18
 kvp: Peak kilovoltage
 mA: Milliampere
 MHz: Megahertz
 mL: Milliliters
 MRI: Magnetic Resonance Imaging
 N/A: Not Applicable
 PET: Positron Emission Tomography
 T1WI: T1-Weighted Imaging
 T2WI: T2-Weighted Imaging
 US: Ultrasound

KEYWORDS

Fibroids; morcellator; seeding; implants; pelvis; uterus; recurrent fibroids; morcellation; Computed Tomography; Ultrasound; leiomyoma

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