

# Leiomyomatosis peritonealis disseminata with formation of endometrial cysts within tumors arising after supracervical laparoscopic hysterectomy

Rozsiana mięśniakowatość otrzewnowa z obecnością torbieli endometrialnych wewnątrz guzów, która wystąpiła po nadszyjkowej histerektomii laparoskopowej

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## Abstract

**Background:** Use of morcellation during laparoscopic hysterectomy may result in seeding of uterine tissue throughout the abdominal cavity and development of 'iatrogenic' leiomyomatosis peritonealis disseminata.

**Case:** Two years after a supracervical laparoscopic hysterectomy a 42-year-old parous women presented with abdominal pain and bloating. CT scans and subsequent surgical exploration revealed multiple solid tumors containing cysts filled with altered blood. Histologically, the tumors had characteristic features of a benign leiomyoma with smooth muscle cells infiltrated by endometrial glands.

**Conclusion:** Pieces of smooth muscle cell and endometrial uterine tissue lost in the abdominal cavity during morcellation may progress to leiomyomatosis peritonealis disseminata with unusual appearance.

Key words: **leiomyomatosis peritonealis disseminata / laparoscopic hysterectomy /**

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## Streszczenie

Zastosowanie morcelacji w celu usunięcia macicy podczas histerektomii laparoskopowej niesie ze sobą ryzyko pozostawienia fragmentów macicy i rozwoju „iatrogennej” rozsiaanej mięśniakowatości otrzewnowej.

**Przypadek:** 42-letnia pacjentka dwa lata po przebyciu nadszyjkowej histerektomii laparoskopowej zgłosiła się z powodu bólów brzucha i wzdęć. W badaniu ginekologicznym stwierdzono obecność guza w miednicy mniejszej. W tomografii komputerowej miednicy i jamy brzusznej oraz podczas laparotomii stwierdzono liczne lite guzy zawierające przestrzenie płynowe wypełnione zhemolizowaną krwią. Badanie histopatologiczne zmian wykazało utkanie charakterystyczne dla mięśniaków gładkokomórkowych macicy oraz obecność gruczołów endometrialnych.

**Wnioski:** Fragmenty mięśnia oraz błony śluzowej macicy pozostawione podczas morcelacji w jamie brzusznej mogą być przyczyną rozwoju rozsiaanej mięśniakowatości otrzewnowej o nietypowej charakterystyce klinicznej.

Słowa kluczowe: **rozsiana mięśniakowatość otrzewnowa / histerektomia laparoskopowa /**

Leiomyomatosis peritonealis disseminata (LPD) is a rare condition characterized by multiple subperitoneal or peritoneal nodules with histologic features of benign uterine leiomyoma. According to the most widely accepted theory LPD lesions growth out of subperitoneal mesenchymal stem cells, which undergo smooth muscle metaplasia under the hormonal stimulation [1]. However, in some cases a link between dissemination of uterine tissue particles during electrical morcellation and development of LPD has been proposed, providing evidence for ‘iatrogenic’ theory of LPD pathogenesis. Presumably, during morcellation pieces of specimens may be dispersed throughout the abdominal cavity, become implanted onto normal tissue and give rise to development of fibrotic nodules and endometrial lesions [1]. Several cases of parasitic leiomyomas, LPD, adenomyosis and endometriosis following morcellation have been reported recently [1, 2].

We present a case of LPD with unusual appearance of tumors distinguished by formation of endometrial cysts within the masses, diagnosed 2 years after a supracervical laparoscopic hysterectomy.

## Case

A 42-year-old women, gravida 2, para 1, presented with vague abdominal pain and bloating suggesting gastrointestinal condition. Prior history revealed a laparoscopic supracervical hysterectomy for symptomatic fibroids performed 26 months earlier. At the time of hysterectomy the largest leiomyoma measured 7.6 x 4.8cm and the weight of removed uterus was 180g. Removal of the tissue specimens was accomplished with electric morcellator. No complications occurred during surgery or during the postoperative period. The follow-up at 1 and 6 months was unremarkable.

The patient underwent also a cesarean section at the age of 29 years, a laparoscopic salpingotomy due to ectopic pregnancy three years later and uterine artery embolization for uterine leiomyomas at the age of 37 years.

On physical examination a firm mobile mass about 6cm in diameter was found in the pelvis on the left side. Pelvic ultrasound demonstrated cystic mass with thick wall and diffuse homogeneous dispersion of low-level echos within it, similar to



**Figure 1.** CT scans demonstrated multiple nodules with regular contours disseminated throughout the abdominal cavity.

observed in the case of endometriomas. CA-125 concentration was elevated at 273.7 IU/L. In view of a suspected ovarian tumor intravenous contrast-enhanced multidetector computed tomography of the abdomen and pelvis was performed.

CT scans revealed multiple well-circumscribed pelvic tumors closely attached to peritoneum and bowel loops (Figure 1).

The largest one measured 8.2cm x 7.8cm x 5.2cm. The density at native scanning varied between 28Hu and 35Hu. Contrast-enhanced images showed heterogenous enhancement of the masses, which presented as thick-walled cystic lesions (Figure 2).

The density of capsules was 79Hu at the arterial phase and up to 105Hu at the venous phase indicating their solid structure, whereas the low-density of central areas (28Hu) suggested accumulation of thick fluid. Additionally, moderate ascites and slightly enlarged paraaortal and iliac lymph nodes were found.

Because of uncertain diagnosis and suspicion of disseminated neoplasm the patient underwent exploratory laparotomy. At laparotomy multiple tumors of varying size were found attached to the peritoneum, omentum and intestines. The ovaries and uterine cervix were normal. The complete resection of lesions as well as trachelectomy and bilateral salpingo-oophorectomy were performed.

Gross appearance of tumors resembled uterine leiomyomas, however at the cross-section they were filled with altered blood. Histologically, the tumors were composed of spindle-shaped muscle cells without signs of malignancy, typical for benign uterine leiomyoma, but in the areas close to cystic lumens smooth muscle cells were infiltrated by endometrial glands (Figure 3).

## Comment

Hysterectomy without laparotomy is now recommended as a procedure of choice for patients requiring removal of the uterus due to benign conditions. Laparoscopic route has been proved to be safe, efficient and cost-effective, and may be regarded as an alternative to abdominal or vaginal hysterectomy [3, 4]. However, some reports published in the recent years have risen the concern that retrieval of tissues by electric morcellation presents a hazard for seeding of uterine or myoma remnants into the peritoneal cavity.

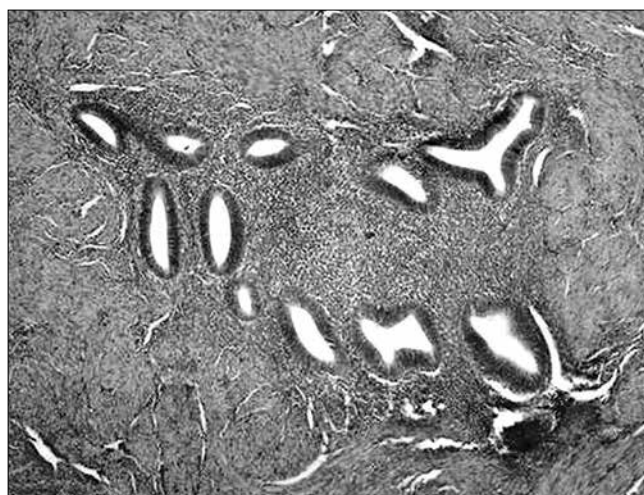
The incidence of implantation and regrowth of retained fragments is largely unknown. Donnez et al. [5] found 8 cases of parasitic tumors following 1450 laparoscopic hysterectomies. In this series, all lesions were composed of myomatous tissue infiltrated by endometrial glands (adenomyosis) giving evidence that both endometrial and smooth muscle components may become implanted. Our case presenting myometrial tumors with internal cysts filled with altered blood and plentiful endometrial glands interspersed between the smooth-muscle cells supports the possibility of regrowth of both components. What's important, these findings imply that there is a risk of dissemination of endometrial cancer or other uterine malignancies if they are not diagnosed before laparoscopic surgery. Such a case has not been presented as yet, however, in a recent report, atypical complex endometrial hyperplasia was found in a mass removed from the pelvis 7 years after laparoscopic hysterectomy [6].

Due to nonspecific symptoms and atypical radiologic features diagnosis of LPD is challenging. On CT scans LPD masses are usually observed as well-circumscribed multiple nodules with contrast-enhancement characteristic to that of myometrium or uterine fibroids. However, when necrosis, degeneration or implantation of endometrial components occur an enhancement is heterogenous, mimicking peritoneal carcinomatosis [7, 8]. Cystic appearance of lesions in our case made preoperative diagnosis even more difficult raising high suspicion of disseminated malignancy.

Reports of 'iatrogenic' LPD related to use of morcellator are scarce. To date, only 2 cases of LPD after laparoscopic hysterectomy and 4 after laparoscopic myomectomy have been presented in the literature [1, 2, 9-11]. Although, the risk of implantation and growth of myometrial and endometrial tissue left in the abdominal cavity after morcellation can be regarded as very low, several important practice measures can be advised to avoid associated serious complications. First of all, it seems reasonable to rule out uterine malignancy before laparoscopic



**Figure 2.** Contrast-enhanced CT showed heterogenous enhancement of the masses with low-density central areas suggestive of accumulation of thick fluid.



**Figure 3.** Histologic findings of 1 of the masses. Oxyphilic spindle-shaped cells without atypia that are characteristic of a uterine leiomyoma are infiltrated by endometrial glands. (magnification x 200).

procedure. Furthermore, during surgery the abdominal cavity should be meticulously inspected and all retained fragments recovered. Finally, a vigorous irrigation should be performed at the end of surgery.

In summary, our case illustrates that use of uterine morcellation may result in seeding of myometrial and endometrial tissue throughout the abdominal cavity that leads to serious morbidity and causes diagnostic dilemma. Although laparoscopic hysterectomy is safe and widely accepted procedure possibility of such a complication should be bear in mind.

**Piśmiennictwo**

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