

## Multiple Iatrogenic Peritoneal Leiomyoma after Laparoscopic Myomectomy and Pregnancy: Case Report

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### Abstract

Peritoneal iatrogenic leiomyomas are a rare condition resulting from cells or small leiomyoma fragments left after laparoscopic morcellation and/or myomectomy. The reported incidence of peritoneal myomas after laparoscopic myomectomy is 0.12 - 1.25%. Steroid exposure (endogenous/exogenous) after laparoscopic morcellation might be a supporting factor for development. Regardless morcellation, gestation and/or enhanced hormonal stimulation are also responsible for peritoneal leiomyomas. Although it is rare, patients should be preoperatively informed about the risk of this condition following laparoscopic surgery. We report a rare case of multiple peritoneal myoma which may have developed from gestation and/or from laparoscopic myomectomy.

**Keywords:** Iatrogenic Peritoneal Leiomyoma; Laparoscopic Myomectomy; Pregnancy

### Introduction

Uterine leiomyoma (fibroids, LM) are the most common benign gynecological condition in women of reproductive age. Laparoscopic myomectomy with and without morcellation is a common gynecologic surgery.

Extrauterine LM can occur as single or multiple benign peritoneal LM (PLM) or as part of a disseminated peritoneal leiomyomatosis (DPLM) after morcellation of LM during myomectomy or endoscopic hysterectomies. Molecular and cytogenetic analyses of these iatrogenic metastatic PLM demonstrate similar chromosomal abnormalities in the PLM nodules as were detected in the original uterine LM [1-3]. Takeda, *et al.* compared the histological features of multiple PLM found 6 years after laparoscopic myomectomy with morcellation with the histological results of the LM removed at initial surgery. The findings were almost identical, and the presence of progesterone-receptors was reported in both the PLM and the previously removed LM [4].

The iatrogenic PLM are often described as parasitic LM [4-7] or iatrogenic parasitic LM [8]. However, parasitic LM are a rare variant of pedunculated subserosal LM that have outgrown their uterine blood supply and become separated from the uterus, receiving blood supply from another source for instance the omentum [9,10].

The first case of PLM after use of the laparoscopic morcellation was reported in 1997 by Ostrzenski [11]. At time there is a fast growing of case reports and reviews regarding pathogenesis and clinics of iatrogenic PLM caused by the seeding of cells or portions of fibroids

during morcellation in case of myomectomy or hysterectomy, especially laparoscopic supracervical hysterectomy [3,4,6,8]. The overall incidence of PLM after laparoscopic morcellation is about 0.12 - 1.25% [3,12]. The use of electric morcellators appears to play a role in this regard [13]. The risk of developing PLM after undergoing electric morcellation is said to range from 0.12 to 0.9% [13,14]. It is not uncommon for the subperitoneal masses to be accompanied by retroperitoneal LM situated at the laparoscopic incision sites after such interventions [11,15,16]. Yanazume, *et al.* published a very rare case with a large peritoneal myoma measuring 12 cm in the superficial adipose tissue under the surgical scar also after laparotomy [17].

PLM may be diagnosed long after laparoscopic morcellation and are often asymptomatic but can present with abdominal or pelvic pain [3]. Hence, this rare condition must be kept in mind whenever a patient presents with abdominal masses following laparoscopic myomectomy or hysterectomy [3].

DPLM is characterized mostly by the presence of numerous smooth muscle nodules or implants beneath the peritoneum. There are two different pathogenetic and clinically forms of DPLM. The first variant occurs during (!) gestation or under (!) extreme hormone exposure including long time use of tamoxifen, and generally regresses once said gestational and/or hormonal environment ceases to be [5,7,18].

The second variant arises as a result of myoma cell dissemination caused in the course of LM surgery, usually in combination with morcellation. This latter variant should be regarded as iatrogenic benign metastatic PLM, and is not associated with special hormone exposure, and can persist beyond menopause. A comprehensive description of DPLM can be found at Köhler, *et al* [19].

Here we present a rare case of a PLD that may be both a pregnancy-associated DPLM and an iatrogenic metastatic PLM.

### Case Report

In a 34-year-old woman a right-sided unclear suprarenal tumor with a diameter of 5 x 7 cm was randomly detected by a CT-scan for other reasons. At that time the patient was without symptoms and was trying to get a second pregnancy. For further diagnostics an MRI was performed (Figure 1, 2 and 5). The MRI confirmed the suprarenal tumor. Additionally, a precaecal tumor with a size of 3 x 2 cm and a uterus with multiple myoma were found.

There were two myomectomies in her history. The first was performed in 1999 by laparotomy revealing a cellular LM. The second one was carried out in 2001 by laparoscopy with morcellement. This time the histological examination revealed an ordinary LM. In addition, she had undergone a cesarean section in 2014 without signs of extrauterine LM.

To clear the suprarenal tumor for safety, a transcutaneous biopsy was performed and revealed a regular LM. Given the history of prior laparoscopic myomectomies multiple ectopic LM that may have induced by pregnancy had been suspected. For this reason, a re-laparoscopy was initiated.

During laparoscopy a double-fist-sized uterus with multiple fibroids was seen. The latter originated from the uterine fundus and were interpreted as primary intramural LM. Both adnexa were completely unremarkable. Additionally, one and two extrauterine fibroids were found in the area of the right and the left tube, respectively. Each was of about 1 cm in size. Further, another 1 cm ectopic fibroid on the right sacrouterin ligament and a pedunculated one on the rectal anterior wall were present. All peritoneal LM were ablated with ultracision. During laparoscopic procedure the ectopic LM on the peritoneum of the caecal region, already detected by MRI, became also apparent.

After elevation of the liver an encapsulated ectopic LM with 5 x 7 cm in size was found, adhering to the posterior wall of the abdomen, but with no direct relation to the liver or kidney, as expected. It was gradually removed with ultracision and deposited in an endobag. After that, the ectopic LM was eliminated from the paracaecal region.

Due to the existing desire for children, the intramural uterine fibroids were also removed by myomectomy. The superficial defect of the uterine fundus was reconstructed by a two-layer, continuous suture with PDS in the sense of a plastic uterine reconstruction.

To avoid spilling, the mandatory morcellation was safely performed with the a large morcellator within in an endobag, with all morcellated fibroids being separately preserved. Thereafter all endobags were removed from the abdomen. Finally, a very thorough flushing of the entire abdominal cavity with several liters of preheated Ringer’s solution using reverse Trendelenburg positioning was carried out until the rinse was completely clear. The final histopathological examination revealed LM with regressively scarred lesions without evidence of cellularity and increased mitotic activity.

Two years after the above procedure, the patient was delivered by a further child by primary cesarean section. There was no evidence of fibroids during the operation.

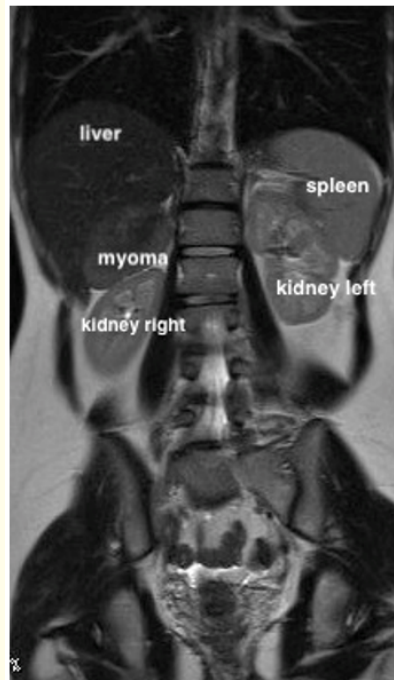


Figure 1: Coronal MRI with an irregular mass of 5 x 7 cm in the area of the right adrenal gland.

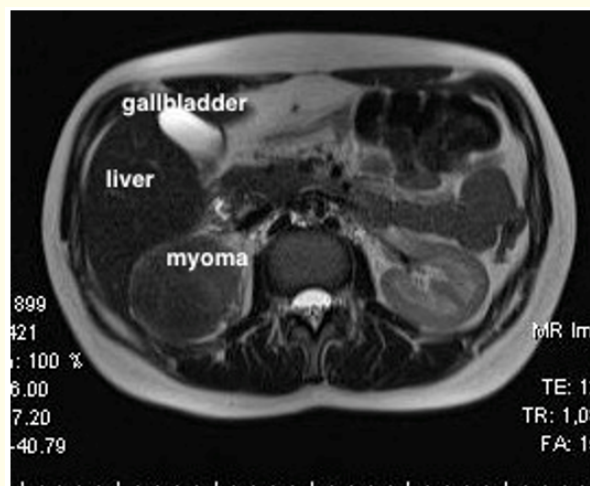
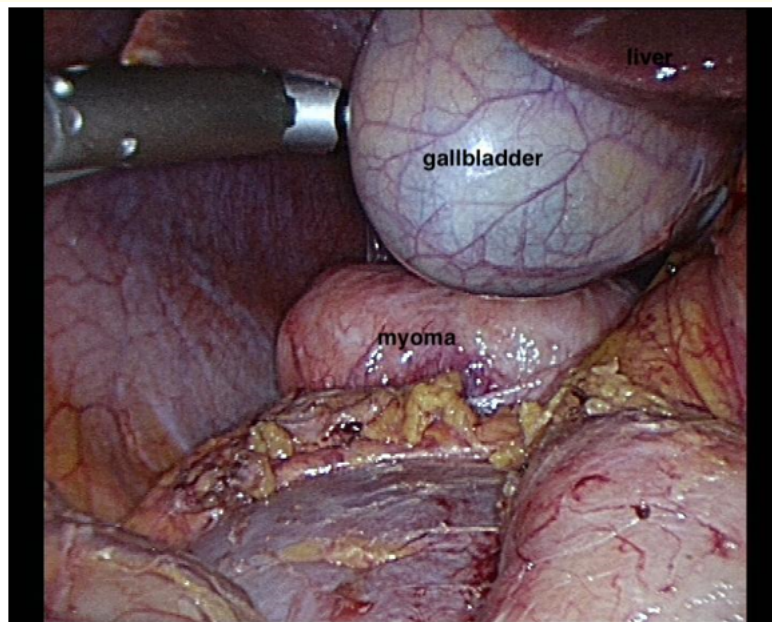
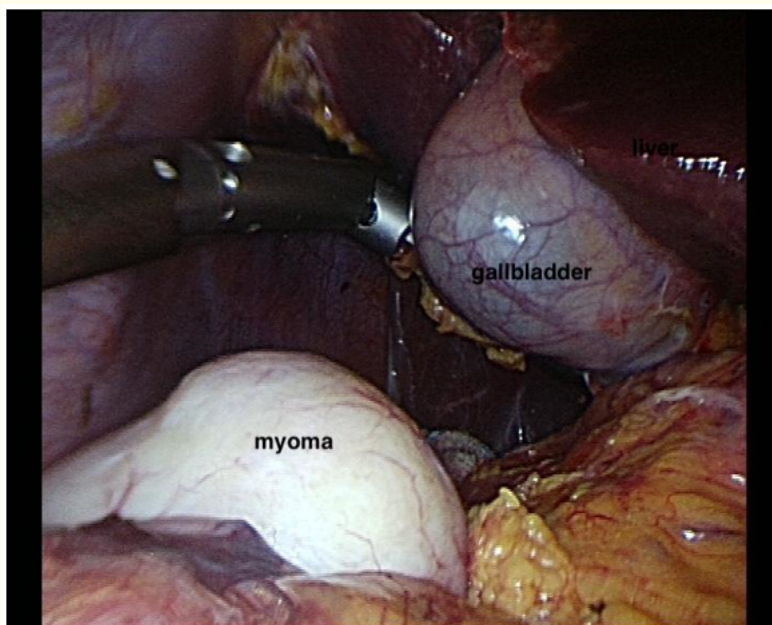


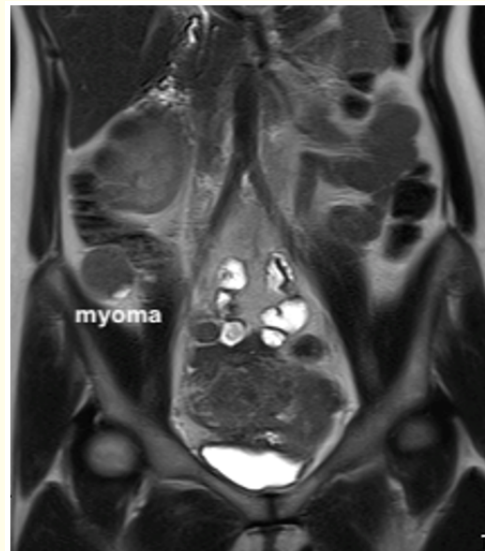
Figure 2: Transverse plane MRI of the tumor from figure 1.



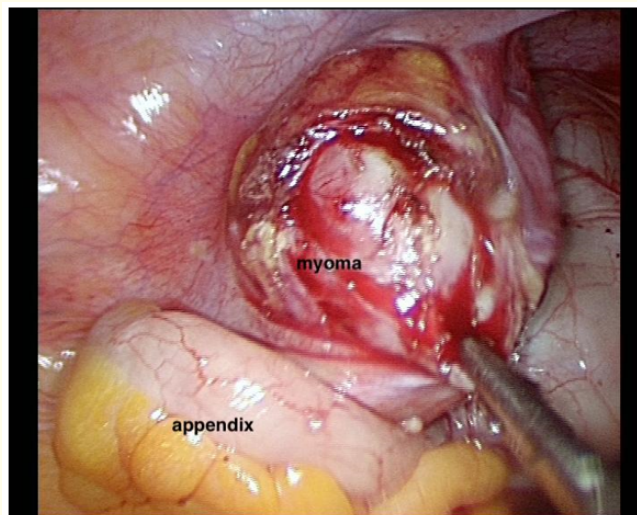
**Figure 3:** Laparoscopic picture of an ectopic leiomyoma of 5 x 7 cm, adhering to the posterior wall and covered by the peritoneum without connections to liver and kidney.



**Figure 4:** Laparoscopic presentation of the ectopic myoma (figure 3) after opening of the peritoneal layer.



**Figure 5:** MRI of a contrast-receiving round tumor of 3 x 2 cm in the caecal region at the right side of the uterus with multiple leiomyoma.



**Figure 6:** Laparoscopic picture of the ectopic leiomyoma from figure 5.

## Discussion and Conclusion

Laparoscopic myomectomy or laparoscopic supracervical hysterectomy with morcellation is a common surgical method in LM and may result in single or multiple PLM with different size due to intraoperative cell spilling. In premenopausal women the further steroid exposure (endogenous/exogenous) or pregnancy after laparoscopic morcellation might be an additional risk factor for development of

this entity [3]. So, Takeda, *et al.* [4] published a case of a woman who was diagnosed with PLM two years after a laparoscopic myomectomy. Over the period of 2 years conservative treatment, the size of the mass remained the same, yet during pregnancy, rapid growth of this mass was observed, supporting the hormonal impact on growth of peritoneal myomas. Cucinella, *et al.* report a case where an asymptomatic PLM was discovered during a caesarean section, 24 months after a laparoscopic myomectomy. The presented own case is somewhat difficult to interpret. Because there were no PLM visible at the first caesarian section and there were no signs of PLM found at the last caesarean section and during the follow up it remains unclear whether the PLM was really pregnancy-associated. According to the available data, the period from primary surgery to the occurrence of PLM ranges from 2 to 30 years [5,3]. It is also occasionally observed that patients with obviously gestation-induced DPLM have previously undergone myoma surgery [20]. In some studies, comprehensive cytogenetic examinations in fact revealed the PLM to have the same genetic attributes as the primarily morcellated LM [2]. Like the primary uterine LM, the PLM lesions also exhibited the same X-chromosome inactivation [21]. Special hormonal exposure in the form of gestation, hormonal contraception or HRT is apparently not required for this type of PLM. In summary we suspect that our patient had late iatrogenic PLM with pregnancies that did not affect this disease.

Whether access is by laparotomy or by laparoscopy, meticulous attention should be paid to basic surgical principles, including attention to complete removal of small fragments of fibroids that may be hidden under bowel or bladder or stuck in port-side cannulas and peritoneal lesions of the abdominal wall. At the end of surgery, the entire abdominal cavity should be flushed with approximately 3 liters of lactated Ringer's solution in upper body elevation, using reverse Trendelenburg positioning until the rinse is completely clear. Furthermore, of the different types of morcellators available, it is important to select the one that provides less tissue scattering [13].

## Conclusion

By using surgical procedures with morcellation, surgeons should be aware of the potential for iatrogenic PLM formation, and intraoperative precautions to minimize occurrence must be the rule. In addition, patients need to be informed about the potential risk of PLM following morcellation.

## Bibliography

1. Miyake T, *et al.* "A case of disseminated peritoneal leiomyomatosis developing after laparoscope-assisted myomectomy". *Gynecologic and Obstetric Investigation* 67.2 (2009): 96-102.
2. Ordulu Z, *et al.* "Disseminated peritoneal leiomyomatosis after laparoscopic supracervical hysterectomy with characteristic molecular cytogenetic findings of uterine leiomyoma". *Genes Chromosomes and Cancer* 49.12 (2010): 1152-1160.
3. Van der Meulen JF, *et al.* "Parasitic myoma after laparoscopic morcellation: a systematic review of the literature". *BJOG: An International Journal of Obstetrics and Gynaecology* 123.1 (2016): 69-75.
4. Takeda A, *et al.* "Rapid growth of parasitic myoma in early pregnancy: previously undescribed manifestation of a rare disorder after laparoscopic- assisted myomectomy". *European Journal of Obstetrics and Gynecology and Reproductive Biology* 162.1 (2012): 117-118.
5. Al-Talib A and Tulandi T. "Pathophysiology and possible iatrogenic cause of leiomyomatosis peritonealis disseminate". *Gynecologic and Obstetric Investigation* 69.4 (2010): 239-244.
6. Sinha R, *et al.* "Parasitic myoma under the diaphragm". *Journal of Minimally Invasive Gynecology* 14.1 (2007): 1.
7. Willson JR and Peale AR. "Multiple peritoneal leiomyomas associated with a granulosa cell tumor of the ovary". *American Journal of Obstetrics and Gynecology* 64.1 (1952): 204-208.

8. Nezhat C and Kho K. "Iatrogenic myomas: new class of myomas?" *Journal of Minimally Invasive Gynecology* 17.5 (2010): 544-550.
9. Brody S. "Parasitic fibroids". *American Journal of Obstetrics and Gynecology* 65.6 (1953): 1354-1356.
10. Clement PB and Young RH. "Atlas of gynaecologic surgical pathology". 3<sup>rd</sup> edition, Elsevier Saunders (2014).
11. Ostrzenski A. "Uterine leiomyoma particle growing in an abdominal- wall incision after laparoscopic retrieval". *Obstetrics and Gynecology* 89.5 (1997): 853-854.
12. Zhang HM., et al. "Non-malignant Sequelae after Unconfined Power Morcellation". *Journal of Minimally Invasive Gynecology* 26.3 (2019): 434-440.
13. Cucinella G., et al. "Parasitic myomas after laparoscopic surgery: an emerging complication in the use of morcellator? Description of four cases". *Fertility and Sterility* 96.2 (2011): e90-e96.
14. Tan-Kim J., et al. "Uterine sarcomas and parasitic myomas after laparoscopic hysterectomy with power morcellation". *American Journal of Obstetrics and Gynecology* 212.5 (2015): 594.e1-10.
15. Moon HS., et al. "Parasitic leiomyoma in the abdominal wall after laparoscopic myomectomy". *Fertility and Sterility* 90.4 (2008): 1201.e1-2.
16. Thian YL., et al. "Leiomyomatosis peritonealis disseminata and subcutaneous myoma -a rare complication of laparoscopic myomectomy". *Abdominal Imaging* 34.2 (2009): 235-238.
17. Yanazume S., et al. "Large parasitic myomas in abdominal subcutaneous adipose tissue along a previous myomectomy scar". *Journal of Obstetrics and Gynaecology Research* 38.5 (2012): 875-879.
18. Heinig J., et al. "Recurrent leiomyomatosis peritonealis disseminata after hysterectomy and bilateral salpingo-oophorectomy during combined hormone replacement therapy". *European Journal of Obstetrics and Gynecology and Reproductive Biology* 111.2 (2003): 216-218.
19. Köhler G., et al. "Variants of leiomyoma (angio- and lipoleiomyoma, cotyledonoid and cellular leiomyoma, leiomyoma with bizarre nuclei, mitotically active, epithelioid and myxoid leiomyoma), smooth muscle tumors with uncertain malignant potential (atypical smooth muscle tumors), disseminated peritoneal leiomyomatosis, benign metastasizing leiomyoma, intravenous leiomyomatosis". In: Köhler G., et al. *Sarcoma of the female genitalia. Volume 1: Smooth muscle and stromal tumors and prevention of inadequate sarcoma surgery.* Walter de Gruyter GmbH, Berlin/Boston (2017): 1-117.
20. Onorati M., et al. "Leiomyomatosis peritonealis disseminata: pregnancy, contraception and myomectomy of its pathogenesis". *Pathologica* 105.3 (2013): 107-109.
21. Bradley J., et al. "Disseminated peritoneal leiomyomatosis clonality analysis by X chromosome inactivation and cytogenetics of a clinically benign smooth muscle proliferation". *American Journal of Pathology* 150.6 (1997): 2153-2166.

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