A case of disseminated peritoneal leiomyomatosis after two laparoscopic procedures due to uterine fibroids

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Abstract
Disseminated peritoneal leiomyomatosis (DPL) is a rare disorder characterized by the presence of multifocal nodules and tumors composed of proliferating smooth muscle tissue, spread throughout the peritoneum. Estrogens and progesterone are considered to be the main factors initiating the formation of disseminated leiomyomatosis. Disseminated peritoneal leiomyomatosis is often asymptomatic, and acyclic vaginal bleeding or pain in the lower abdomen is associated with leiomyomatosis rebuilt uterus corpus. Disseminated peritoneal leiomyomatosis can have other ambiguous presentation. The difficulty in DPL diagnosis is that it is not always accompanied by scattered leiomyomas and can occur after menopause. Some cases of DPL are associated with surgical procedures on uterine fibroids, especially with the use of a morcellator. We present the case of a 39-year-old woman with DPL who underwent laparoscopic myomectomy and laparoscopic supracervical hysterectomy before the final diagnosis of DPL. After the complete surgical treatment performed in our center the patient is free of symptoms.

Key words: laparoscopy, uterine fibroids, disseminated peritoneal leiomyomatosis, power morcellation.

Introduction
Disseminated peritoneal leiomyomatosis (DPL) is a rare disorder characterized by the presence of multifocal nodules and tumors composed of proliferating smooth muscle tissue, spread throughout the peritoneum. The first case of DPL was described in 1952 [1]. To date, approximately 150 cases have been described [2]. The disease is mainly asymptomatic; only a few cases of abdominal pain, urinary tract problems and vaginal or rectal bleeding have been described [3–5]. Fibroblasts and myofibroblasts arising along the submesothelial tissue located in the pelvis may be the primary focus of leiomyomatosis [3]. Estrogens and progesterone are considered as the main factors initiating the formation of disseminated leiomyomatosis. Their receptors can be revealed by immunohistochemical methods [6]. The difficulty in DPL diagnosis is that it is not always accompanied by scattered leiomyomas and can occur after menopause [5].

We present the case of a 39-year-old woman with DPL who underwent laparoscopic myomectomy and laparoscopic supracervical hysterectomy before the final diagnosis of DPL. After the complete surgical treatment performed in our center the patient is free of symptoms.

Case report
The 39-year-old multipara was admitted to hospital due to a tumor-like change, located in the proximity of the cervical stump, found during a transvaginal ultrasound control scan. In 2011 the patient underwent laparoscopic fibroid enucleation surgery. In 2013 she underwent laparoscopic amputation of
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The uterine corpus due to numerous fibroids. In both cases fibroid morcellation was performed. The patient denied pain, constipation and nausea. Transvaginal ultrasound scan revealed a mobile, solid tumor with regular shape and size of 83 × 61 × 60 mm. Magnetic resonance imaging of the abdomen and pelvis revealed a heterogeneous, richly vascularized tumor of the same size as mentioned above. Increase in size of internal iliac lymph nodes was an additional magnetic resonance imaging (MRI) finding. The patient’s laboratory tests were within the normal range. Due to the suspicion of a proliferative process the clinical team decided to perform laparotomy surgery.

A tumor 8 × 7 × 6 cm in size coming out of a peritoneum was found during surgery. Multiple smaller tumors 3 cm in size coming out of the peritoneum, intestinal appendicles, pelvic ligaments and bladder wall were also present; on both sides ovaries were unchanged. The operating team decided to remove all the changes from the peritoneum, the bowel mesentery and omentum as well as to remove the cervix and both ovaries (Photos 1 and 2). Bilateral lymphadenectomy was also performed. Histopathological examination revealed leiomyomatic tissue (Photos 3–6). The patient was discharged home on the fifth day after surgery in good condition. After 6 months the patient does not report any gynecological ailments.

Discussion
Disseminated peritoneal leiomyomatosis is a rare disorder characterized by the presence of multifo-
cal nodules arising from the proliferation of smooth muscle cells, fibroblasts and myofibroblasts emerging along submesothelial tissue located in the pelvic cavity [7, 8]. Disseminated peritoneal leiomyomatosis affects women in reproductive age and is often revealed during caesarean section [7]. As mentioned above, there are cases of DPL in postmenopausal women. In many patients disseminated tumors are accompanied by a leiomyomatous uterine corpus. Robles-Frías et al. described a case of DPL affecting the uterus, which corresponded in size to 16–18 weeks of gestation [9].

Abnormal submesothelial tissue sensitivity to ovarian hormones may also be one of the causes of DPL. That is why most of the DPL cases are described in patients who are pregnant or use oral contraceptives [2, 5, 10]. The evidence for these theories is as follows: DPL is only diagnosed in women, especially pregnant or on oral contraception; gonadectomy causes the resolution of tumors; estrogen and progesterone receptors are found in tumor tissues [2, 6, 11, 12]. Disseminated peritoneal leiomyomatosis occurrence in postmenopausal women can be explained by hypersensitivity of pathological tissue receptors under the influence of correct hormone concentrations [13].

Disseminated peritoneal leiomyomatosis is often asymptomatic, and acyclic vaginal bleeding and pain in the lower abdomen are associated with leiomyomatous rebuilt uterus corpus. Disseminated peritoneal leiomyomatosis can have other ambiguous presentation. Some cases of DPL are associated with surgery of uterine fibroids, especially using a power morcellator. Morcellation is a risk factor of DPL presentation [14, 15]. One author describes fibroid tissue implantation at the point of laparoscopic trocar entry after laparoscopic myomectomy; the patient was referred because of the easily palpable tumor [16]. In our patient the definitive diagnosis was preceded by both laparoscopic myomectomy and laparoscopic amputation of the uterus. Both fibroid and uterine corpus were extracted from the peritoneal cavity using a power morcellator. This could involve the risk of leaving small fibroid fragments in the abdominal cavity, which continued progression under the influence of patients’ sex hormones.

Final DPL diagnosis can only be established through a histopathological examination and finding of smooth muscle cells without atypia or necrosis. Differential diagnoses are as follows: parasitic leiomyoma, intravenous leiomyomatosis, peritoneal carcinomatosis or leiomyosarcoma [5, 7, 17]. Leiomyomatosis must always be differentiated from metastatic leiomyosarcoma. It is believed that leiomyosarcoma nodules are generally fewer in number, bigger and they infiltrate surrounding tissues [5].

Disseminated peritoneal leiomyomatosis treatment methods include surgical resection of leiomyomatous tumors [2, 18]. Other options are pharmacological treatment and hormonal or surgical castration [19]. There are attempts to use chemotherapy in cases of DPL [20]. It is currently believed that laparoscopic morcellation can contribute to relapse of leiomyomatous changes [14, 21, 22]. After the FDA alarm was exposed, a fierce debate about continuation of morcellation use started, and it still
takes place; experts are divided [15]. Some authors describe the presence of leiomyomatosis in women after laparoscopic myomectomy. It is explained that fibroid tissue fragments which are left after laparoscopy in the abdominal cavity are rising due to stimulation by steroid hormones. Therefore, in the case of recurrent fibroids, some authors suggest bilateral oophorectomy to eliminate the main source of estrogens [5, 10, 14]. Therefore, in case of recurrent fibroids or leiomyomatosis, some authors suggest ovariectomy in order to eliminate the main source of estrogens [5, 23].

GnRH agonists and aromatase inhibitors can be an alternative to surgical treatment. Lewis et al., based on an analysis of 5 cases, found GnRH agonists and aromatase inhibitors as the optimal treatment for inoperable, multifocal changes, and for women who want to avoid fibroid surgery [24]. One author describes the successful use of ulipristal acetate for the treatment of metastatic leiomyomas [25]. The pharmacological approach is an interesting option and appears to be very effective. It must be remembered that in the case of drug-only treatment, there is a lack of histopathological verification (if the process is not malignant). The combined approach of surgery with subsequent pharmacological therapy aimed at reduction or extinction of changes may be a new approach to the DPL problem.

Conclusions

Disseminated peritoneal leiomyomatosis is a very rare disease. It requires differentiation from other peritoneal tumors for the best solution choice. Laparoscopy on uterine fibroids may be a cause of DPL occurrence, particularly in the case of incautious morcellation. Surgery remains a mainstay of treatment when hormone therapy can be offered for patients who do not agree to undergo surgery or are in a high stage or severity of the DPL disease.

Conflict of interest

The authors declare no conflict of interest.

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