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CASE REPORT

# Leiomyomatosis Peritonealisis Disseminata Resembling Intra-Abdominal Malignancy

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Leiomyomatosis peritonealisis disseminata (LPD) is a rare benign tumor characterized by the dissemination of smooth muscle-like cell nodules within the peritoneal cavity. Diagnosing LPD is challenging for clinicians due to its nonspecific clinical presentation and its dissemination throughout the peritoneal cavity, which mimics intra-abdominal malignancy. In the present study, we present the case of a 47-year-old woman with severe abdominal pain and fever initially diagnosed with intra-abdominal abscess or peritoneal carcinomatosis. Exploratory laparotomy was performed to resect several leiomyoma-like tumors in the subperitoneal space. LPD was confirmed histopathologically.

Key words: Leiomyomatosis peritonealisis disseminata, carcinomatosis, leiomyoma

## INTRODUCTION

Uterine leiomyomas are the most common benign gynecologic neoplasms among women, 20%–25% of whom are diagnosed with uterine leiomyoma when they are of reproductive age.[1] Leiomyomatosis peritonealisis disseminata (LPD) is a rare benign tumor characterized by the dissemination of smooth muscle-like cell nodules in the peritoneal cavity. LPD was first described by Kelly and Cullen in 1909, and the first case was reported by Willson and Peale in 1952.[2] This disease was designated as LPD by Taubert *et al*. in 1965.[3]

Only approximately 200 cases have been reported in the literature to date. LPD is observed more often in premenopausal women rather than in postmenopausal women.

Its etiology remains unclear. Some hypotheses have been proposed, including a history of iatrogenic origin, such as previous myomectomy with leiomyoma fragments and peritoneal metaplasia activated by steroid hormones.[4]

Clinicians find diagnosing LPD to be challenging due to its nonspecific clinical presentation, dissemination throughout the peritoneal cavity, and similarities to intra-abdominal or peritoneal carcinomatosis. We present a case of LPD that was initially diagnosed as intra-abdominal carcinomatosis.

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## CASE REPORT

A 47-year-old Taiwanese G0P0 woman had a history of three myomectomies due to recurrent leiomyoma of the uterus. She first presented with fever and abdominal pain and was admitted to a suspected liver abscess. Physical examination revealed significant tenderness of the lower abdomen with no peritoneal signs or rebounding pain. Laboratory data revealed leukocytosis (10,370/μL), elevated serum C-reactive protein levels (24.68 mg/dL), and *Escherichia coli* bacteremia. Computed tomography revealed an 8.6 cm × 8.5 cm × 5.1 cm heterogeneous mass occupying the right hepatic lobe and a 7.1 cm × 8.0 cm × 4.1 cm heterogeneous mass in the right lower abdomen, which was attached to the ascending colon, [Figure 1] favoring a diagnosis of intra-abdominal abscess or metastatic tumor. Elevated serum cancer antigen 125 (CA-125) levels were also noted (235.1 U/mL). Due to persistent symptoms despite treatment with advanced antibiotics and suspicion of gynecologic malignancy with carcinomatosis, biopsy of the hepatic tumor was performed, and pathology revealed reactive hepatitis with focal bridging fibrosis. Colonoscopy revealed no neogrowth in the colon. Exploratory laparotomy was



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performed due to persistent abdominal pain, and 7-cm and 6-cm masses over the right subdiaphragm with blood supply from the peritoneum and over the right lower quadrant, lateral to the ascending colon, with blood supply from the mesentery were noted intraoperatively [Figure 2]. The connection between the masses and surrounding organs was thin and loose. Subtotal hysterectomy and debulking of the tumors were performed smoothly, and pathology showed leiomyomas [Figure 3]. The patient recovered uneventfully with abdominal pain, and the fever subsided gradually. The patient was discharged a week after the operation, and a fair recovery was reported during later visits for a year.

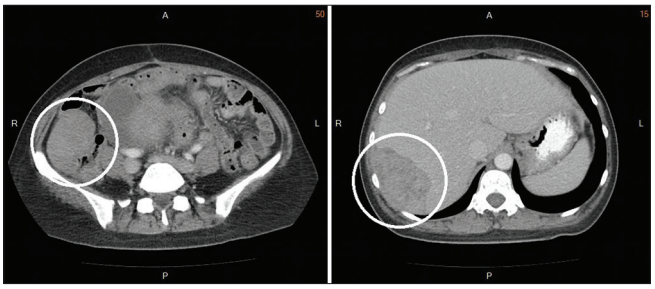

**Figure 1:** A 8.6 cm × 8.5 cm × 5.1 cm heterogeneous mass occupying the right hepatic lobe (left) and 7.1 cm × 8.0 cm × 4.1 cm heterogeneous mass over right lower abdomen attached to the ascending colon (right) are visible on computed tomography. These features resemble those of intra-abdominal abscess or metastatic tumors

## DISCUSSION

Leiomyomas are benign tumors that occur in women of reproductive age. They consist of smooth muscle and connective tissue and are usually found in the genitourinary tract. Leiomyomas in unusual locations with unusual growth patterns are classified as intravenous leiomyomatosis, benign metastasizing leiomyoma, or LPD.[5]

LPD is a rare benign neoplasm characterized by the dissemination of smooth muscle-like cell nodules in the peritoneal cavity. Fewer than 200 cases have been reported to date and the possible pathophysiology of LPD remains unclear. The potential hypotheses are categorized as hormonal, genetic, and iatrogenic theories. According to the hormonal theory, estrogen secretion causes metaplasia of mesenchymal cells into myoblasts, myofibroblasts, and fibroblasts. Regarding the genetic theory, some researchers have suggested that chromosomal abnormalities contribute to the implantation and proliferation of benign smooth muscle tissue. LPD after laparoscopic myomectomy rather than myomectomy or hysterectomy started becoming increasingly common, which supports the iatrogenic theory.[5,6] The possible mechanism is that pieces of myoma may be left behind during morcellation to remove it from the abdominal cavity.

The diagnosis of LPD is challenging. Differential diagnosis of LPD includes peritoneal carcinomatosis, disseminated leiomyosarcoma, and gastrointestinal tract tumors. It is usually asymptomatic or presents with nonspecific symptoms, such as abdominal pain and distension. Ultrasonography, computed tomography, and magnetic resonance imaging, which reveal numerous well-defined nodules with smooth surfaces in the peritoneal cavity can be used to diagnose LPD.[6,7] However, the disseminated pattern of leiomyomas is difficult to distinguish from peritoneal carcinomatosis, and LPD can be mistaken for enlarged lymph nodes when it is located close to the vessels. Tumor markers such as CA-125, CA19-9, and carcinoembryonic antigen (CEA) may not help to exclude possible malignancies because LPD reportedly sometimes causes elevated tumor marker levels.[7]

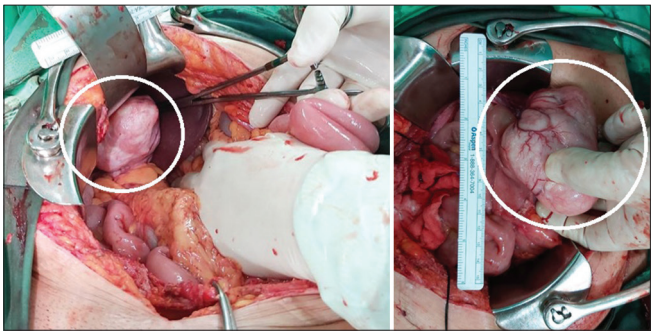

**Figure 2:** A 7-cm mass located over right subdiaphragm (left) and 6-cm mass located over right lower quadrant lateral to the ascending colon with blood supply from the mesentery artery (right) can be seen intraoperatively

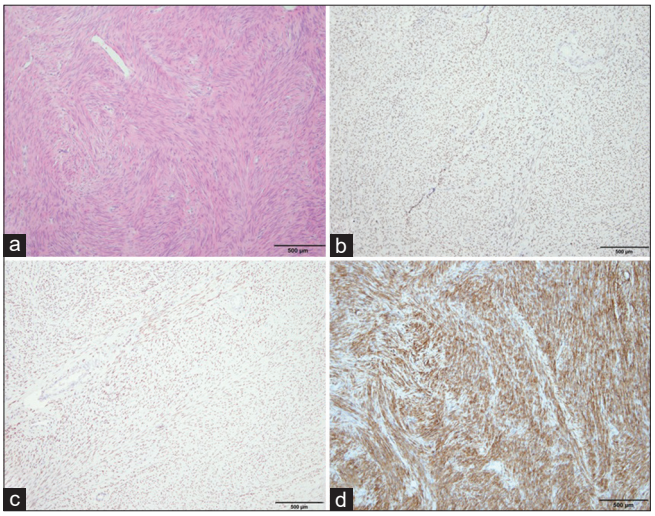


**Figure 3:** (a) Histological examination of resected specimens: fibrous and muscular tissue with the presence of spindle-shaped cells (hematoxylin–eosin staining, ×100), immunohistological staining shows reaction for estrogen receptors (b), progesterone receptors (c), and smooth muscle actin (d)

A definite diagnosis of LPD should be confirmed through histopathological examination. Macroscopically, there

are multiple diffuse nodules on the peritoneal surface that can range from rubbery to hard. Microscopically, LPD resembles uterine leiomyomas. Microscopy shows smooth muscle nodules without atypia. Fibroblasts, myofibroblasts, decidual cells, and sporadic endometrial stromal cells may be noted.[7,8]

There is no consensus or guidelines for the management of LPD. Management should be individualized according to the patient's age, desire for conception, and severity of symptoms. Conservative treatment such as hormone therapy is preferred for patients that desire conception. If surgery is necessary, debulking and omentectomy along with hormone therapy are recommended. For menopausal patients or patients who do not desire conception, debulking, total hysterectomy with bilateral salpingo-oophorectomy, and omentectomy may be the best alternative.[6,9] For symptomatic cases like this patient, surgical intervention with tumor debulking gives rise to symptomatic relief and good outcomes.

## CONCLUSION

LPD is a rare benign disease with diffuse leiomyomas in the subperitoneal space that mimics carcinomatosis. Differentiating LPD from peritoneal carcinomatosis, disseminated leiomyosarcoma, and gastrointestinal tract tumors is challenging. Decision-making for its management depends on the patient's age, desire for conception, and severity of symptoms. Rare cases of recurrence and malignant transformation of LPD have been reported.[10] Patients should thus be closely monitored after the intervention.

### Ethical approval

The Institutional Review Board of Tri-Service General Hospital approved the study (TSGHIRB: C202105193). All of the patients enrolled in the study were agreed to participate and signed the consent form. The researcher ensured patient data confidentiality by storing the data in a file that were required a password to access. Personal identification was by no means included in the data file, and also assured to the patients that their data would not be used for purposes other than this research.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

## REFERENCES

1. Payson M, Leppert P, Segars J. Epidemiology of myomas. Obstet Gynecol Clin North Am 2006;33:1-11.
2. Willson JR, Peale AR. Multiple peritoneal leiomyomas associated with a granulosa-cell tumor of the ovary. Am J Obstet Gynecol 1952;64:204-8.
3. Taubert HD, Wissner SE, Haskins AL. Leiomyomatosis peritonealis disseminata; an unusual complication of genital leiomyomata. Obstet Gynecol 1965;25:561-74.
4. Yoshino Y, Yoshiki N, Nakamura R, Iwahara Y, Ishikawa T, Miyasaka N. Large leiomyomatosis peritonealis disseminata after laparoscopic myomectomy: A case report with literature review. Int J Surg Case Rep 2020;77:866-9.
5. Vaquero ME, Magrina JF, Leslie KO. Uterine smooth-muscle tumors with unusual growth patterns. J Minim Invasive Gynecol 2009;16:263-8.
6. Al-Talib A, Tulandi T. Pathophysiology and possible iatrogenic cause of leiomyomatosis peritonealis disseminata. Gynecol Obstet Invest 2010;69:239-44.
7. Thang NM, Thien DH, Huyen Anh NT, Cuong TD. Leiomyomatosis peritonealis dissemianata five years after laparoscopic uterine myomectomy: A case report. Ann Med Surg (Lond) 2021;66:102377.
8. Bekkers RL, Willemsen WN, Schijf CP, Massuger LF, Bulten J, Merkus JM. Leiomyomatosis peritonealis disseminata: Does malignant transformation occur? A literature review. Gynecol Oncol 1999;75:158-63.
9. Soni S, Pareek P, Narayan S. Disseminated peritoneal leiomyomatosis: An unusual presentation of intra-abdominal lesion mimicking disseminated malignancy. Med Pharm Rep 2020;93:113-6.
10. Ferrario L, Zerbi P, Angiolini MR, Agarossi A, Riggio E, Bondurri A, *et al.* Leiomyomatosis peritonealis disseminata: A case report of recurrent presentation and literature review. Int J Surg Case Rep 2018;49:25-9.