

### **Case Report**

# Disseminated peritoneal leiomyomatosis: A case report<sup>☆</sup>

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#### ABSTRACT

Disseminated peritoneal leiomyomatosis (DPL) or leiomyomatosis peritonealis disseminata is a sporadic benign disease characterized by several solid peritoneal smooth muscle nodules that proliferate along the abdominopelvic cavity. The source of the condition is undetermined, although suspected causes include iatrogenic and hormonal stimulation. It primarily affects women of reproductive age. Imaging investigations are important in determining the extent of lesions and the presence of malignancy. There are no conventional therapeutic guidelines for the therapy of DPL, hence the risk of malignant transformation is low. We discuss the case of a 41-year-old woman who had a previous laparoscopic hysterectomy and presented 4 years later with numerous peritoneal tumors the diagnosis of DPL was suspected by computed tomography and magnetic resonance imaging, and confirmed by histology.

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#### Introduction

Disseminated peritoneal leiomyomatosis (DPL) is an uncommon but benign entity defined by the development of several smooth muscle nodules of varied sizes dispersed throughout the peritoneal cavity, simulating a malignancy but displaying benign histological findings [1].

#### **Case presentation**

A 41-year-old female, unmarried, with irregular menstrual history, no co-morbidities, without a history of using oral contraceptives or family history of any neoplasm, had surgery for uterine leiomyoma 4 years ago. The patient was presented with complaints of recurrent abdominal discomfort. An

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Fig. 1 – Abdominal CT scan reveal multiple well-defined lobulated solid mass in abdomen, pelvis, peritoneal cavity, in bilateral iliac fossa (A) before contrast and after contrast (B) and (C).

abdominal ultrasound was performed and found a large peritoneal mass adhering to the anterior abdominal wall reaching the umbilicus with no evidence of ascites or liver metastases, and the rest of the sonographic examination was unremarkable. Contrast-enhanced computed tomography (CT) was suggestive of hypodense well-defined lobulated solid masses in the abdomen, pelvis, and peritoneal cavity in the bilateral iliac fossa, left lumbar region, and pouch of Douglas, with intense and homogenous enhancement, responsible for bilateral ureterohydronephrosis (Fig. 1).

Abdominal magnetic resonance imaging (MRI) revealed large, mildly enhancing, predominantly solid masses that fill the pelvis and extend into the lower abdomen, hypo intense in T1-weighted images, iso intense on T2-weighted images, and homogenous enhancement in dynamic contrast enhancement images. No ascites or omental caking is present and there is no metastatic disease demonstrated in the liver or osseous structures (Fig. 2).

Blood laboratory testing revealed a slightly elevated CA 19-9 and CA-125, a negative AFP (Alpha-fetoprotein and Beta HCG (Human chorionic gonadotropin). Sonographically guided biopsy of the anterior abdominal wall mass revealed only smooth muscle and confirmed the diagnosis of DPL. Histological examination of peritoneal nodule shows proliferation made of long beams cut at right angles and cells have abundant eosinophilic cytoplasm, nonatypical elongated nuclei with low mitoses. Without any sign of malignant cells or lymphoid tissue (Fig. 3).

#### Discussion

DPL is a rare benign entity characterized by numerous peritoneal nodules that frequently resemble peritoneal carcinomatosis. It is thought to be caused by a metaplasia of submesothelial multipotent mesenchymal cells. Less than 150 cases of DPL have been recorded in English-language literature since it was first characterized by Wilson and Pearl in 1952. The disease's asymptomatic nature may explain the disease's underestimated incidence of 1/10.000.000 [2]. The pathogenesis of DPL is unknown, but several theories have

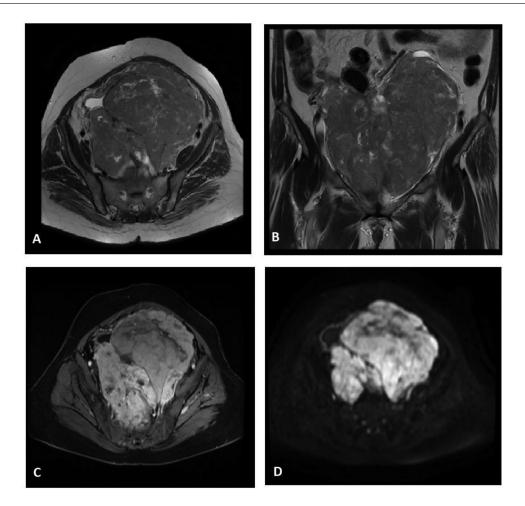


Fig. 2 – Lower abdominal magnetic resonance imaging showing large masses. The signals were not uniform, and the boundaries were clear on T2-weighted images (A), and homogenous enhancement in dynamic contrast enhancement images (C); (A) axial view, (B) coronal view, (D) DWI show diffusion restriction.

been proposed, including the hormonal hypothesis, which states that high levels of exogenous or endogenous female gonadal steroids can stimulate smooth cell differentiation, and is supported by the fact that DPL is more common in women of reproductive age, following the administration of oral contraceptives, or during pregnancy [3]. It might also be induced after myoma morcellation during laparoscopy, with the risk of tumor debris leaking into the abdominal cavity; a recent article emphasized the link between laparoscopic myomectomy and the incidence of DPL (0.12%-0.95%) [4], which is the situation with our patient. Although this condition is benign and has an excellent outcome, there is a 2%-5% chance of neoplastic transformation [5]. Before establishing any therapeutic approach, imaging investigations must be performed to assess the size of the lesions, the existence any sign of malignancy, and to distinguish this largely benign illness from other threatening peritoneum disorders.

DPL imaging findings include several, firm and solid, complicated soft-tissue peritoneal lesions that resemble typical uterine leiomyomas. They are detected on ultrasonography as homogeneously hypoechoic nodules with well-defined wall and a smooth surface. Contrast-enhanced computed tomography CT reveals hypodense, well-defined firm nodules with significant and uniform enhancement, whereas the surrounding peritoneum is usually normal [6]. However, when necrosis, degeneration, or endometrial implantation occurs, the enhancement is heterogeneous, simulating peritoneal carcinomatosis [7]. On magnetic resonance imaging, they appear as well-defined solid masses that are hypo-intense on T1weighted images, hypo-intense, iso-intense, or hyper-intense on T2-weighted images depending on the tumor's water content [8], and consistent and uniform enhancement on dynamic contrast enhancement images. DPL exhibiting malignant transition displays necrosis and hemorrhage in the masses. When DPL or uterine leimyosarcoma is accompanied by hyperintense T2-weighted MR images and diffusion restriction, an unusual histological type of leiomyoma with high cellularity should be investigated [9]. In our case, despite the diffusion restriction, there was any sign of malignancy in the histological examination.

The main differential diagnoses for DLP are peritoneal carcinomatosis, disseminated leimyosarcoma, and endometriosis.

There is not a recognized DPL management protocol. The therapeutic strategy depends on the patient's age, the clinical features, and the desire for conception. Asymptomatic DLP

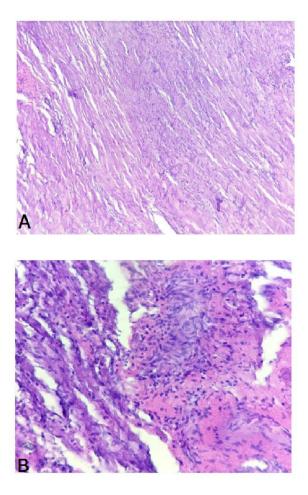


Fig. 3 – Histological examination of peritoneal nodule shows (A) proliferation made of long beams cut at right angles (Hx100) (B) cells have abundant eosinophilic cytoplasm and non-atypical elongated nuclei with low mitoses (Hx400).

requires no therapy, Surgical excision is recommended if the patient is symptomatic with a persistent or recurring lesion that is not responding to hormone treatment or if the patient refuses surgical castration [5].

#### Conclusion

DPL is a rare benign disease mimicking disseminated peritoneal malignancy. It should be suspected in a patient with an intraperitoneal abdominopelvien mass or masses, particularly if the patient has a history of fibroid uterus or has had a hysterectomy for a uterine leiomyoma. It is frequently misdiagnosed due to its rarity and potential lack of familiarity.

#### Author contributions

All authors contributed to this work. All authors have read and approved the final version of the manuscript.

#### Patient consent

Written informed consent for publication was obtained from patient.

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