DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20232315

Case Report

Disseminated peritoneal leiomyomatosis-diagnostic dilemma in an acute presentation-a case report

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Received: 25 June 2023 Accepted: 18 July 2023

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ABSTRACT

Diffuse peritoneal leiomyomatosis is a rare benign condition which has a multifactorial origin with genetic or hormonal component leading to metaplasia of peritoneal mesenchymal cells. Thus, a combination of radiology with clinical correlation is an ideal approach for diagnosis. Though benign, radiologically it could give a picture of malignancy. In our case, patient came with an acute presentation resembling torsion ovary which usually needs emergency detorsion. In this scenario, multiple radiology component directed the case towards malignancy while ultimately a history, clinical correlation and biopsy revealed its benign nature. Reviewing the reported cases of disseminated peritoneal leiomyomatosis (DPL) incidence has slightly increased in recent years and is more common in patients with a past history of unconfined laparoscopic myomectomy wherein spillage of the myoma content into the abdominal cavity can trigger DPL as these are hormone sensitive tissues. The indolent course of the disease usually suggests a borderline disease, but only histological and immunohistochemical studies can confirm DPL, showing smooth muscle cells without nuclear atypia, strongly expressing estrogen and progesterone receptors. Mainstay of treatment targets in curbing the hormone influence on DPL and surgical management.

Keywords: Diffuse peritoneal leiomyomatosis, Estrogen, Progesterone receptors

INTRODUCTION

Disseminated peritoneal leiomyomatosis (DPL) is a rare condition. It is also known as leiomyomatosis peritonalis disseminate (LPD) and is characterized by the presence of multiple leiomyomas throughout the peritoneal cavity. The first case was reported by Willson and Peale in 1952, however, the entity was clearly described and named by Taubert et al in 1965.¹ DPL arises as a result of metaplasia of sub-peritoneal mesenchymal stem cells to smooth muscle, fibroblasts, myofibroblasts and decidual cells under the influence of estrogen and progesterone.² Secondary implantation of myoma into the abdominal cavity appears as a small peritoneal nodule and hence preoperative diagnosis of DPL is challenging and it can be

confused with disseminated intra-abdominal malignancy. There are no specific methods to diagnose DPL in the preoperative period.

Recently, the incidence of parasitic leiomyomas and DPL has increased comparatively after the introduction of laparoscopic power morcellation. With the increasing demand for minimally invasive surgeries, the need for morcellation has increased which also includes morcellation induced iatrogenic pathologies.

We report a case which was evaluated for acute pain with abdominal mass with a past surgical history of laparoscopic myomectomy and her course of diagnosis and management.

CASE REPORT

A 41 years old female presented to the ER at night with complaints of severe left sided lower abdominal pain for the past 4 days which was not relieved with medications and was associated with nausea. On admission, she had high temperature, palpitation and severe anemia (Hb-5 gm/dl).

She has two living children through cesarean sections and has undergone concurrent sterilization.

She had undergone lap myomectomy 5 years ago in the same institution for intramural fibroid (8×8 cm) in which the myoma was removed by uncontained power morcellation. Post op recovery was uneventful.

As per the patient, she was apparently normal 1 year back after which she had heavy menstrual bleeding, for which ultrasound pelvis was done at that time which showed two fibroids in the uterus, largest measuring 5×5 cm and a unilocular simple right ovarian cyst measuring 5×6 cm. She lost further follow up in view of covid pandemic.

On the day of admission, she presented with acute abdominal pain with tender mobile abdominal mass of 18-20 weeks size with fever and severe anaemia. Since she presented with acute abdominal pain and fever with an ultrasound finding taken a year back with ovarian cyst, a provisional diagnosis of ovarian torsion was made. Ultrasound abdomen was done which revealed solid mass lesion in left iliac region with preserved ovarian blood supply and thus torsion of ovarian cyst was ruled out. Since the diagnosis was not clear, general surgery opinion was sought and as per their suggestion, MRI abdomen was done which revealed an irregular lobulated mass lesion in left iliac fossa region between descending colon and jejunum of 7.8×6.1×4.6 cm size with significant peritoneal stranding. Both ovaries were found adherent to uterus with few small functional cysts. Uterus was enlarged with submucous fibroid of 6.9×5.3 cm. A differential diagnosis of para-ovarian mass or mesenteric mass lesion was made for the left iliac fossa mass. In the meantime, her temperature normalised and abdominal pain got settled. Two units of packed RBCs were transfused to correct anaemia. The possibility of gastro intestinal stromal tumour (GIST) was raised and whole-body positron emission tomography-computed tomography (PET-CT) was sought to rule-out malignancy, which revealed multiple solid lesions ranging from 2×2 to 7×10 cm with minimal metabolic activity, suggesting the possibility of disseminated endometriosis or leiomyomatosis and there was a solid lesion $(2.1 \times 1.5 \text{ cm})$ in the left lateral abdominal wall. Ultrasound guided biopsy of the abdominal wall mass lesion was done which revealed the lesion to be of spindle cell origin without atypia suggesting the possibility of leiomyoma. With the past history of laparoscopic myomectomy with uncontained power morcellation, preop diagnosis of DPL was made. Diagnostic laparoscopy was done and after confirming the diagnosis, proceeded

with laparoscopic hysterectomy with excision of 4 myoma attached to the omentum and small intestine which were retrieved vaginally. Excision of the anterior abdominal wall leiomyoma was done simultaneously. Histopathologic examination of the sample confirmed the diagnosis.



Figure 1: Laparoscopic finding of fibroid over omentum.



Figure 2: Laparoscopic finding of fibroid over small intestine.

DISCUSSION

Preoperative diagnosis of DPL is challenging because patients are often asymptomatic or present with atypical symptoms such as abdominal pain and discomfort, and irregular vaginal bleeding. In contrast to the case reported by Julien et al, DLP was diagnosed in the post-menopausal age group in their case with high chance of recurrence.¹ In their case, further suppression of DLP was provided by hormonal suppression with OCP. As in our case, the patient was asymptomatic for 4 years post-myomectomy and presented with DLP within 5 years of laproscopic myomectomy. DLP's usually presented within 10 years of power morcellation. As reported by Xiao et al with the advent of minimally invasive surgery like laparoscopic myomectomy with morcellation, the incidence of DPL has further increased.⁴ However, both cases showed similarity in having a common differential diagnosis of GIST and a complete pre-operative course directing us towards

malignancy, wherein a final histopathology confirmed the diagnosis. DPL being a rare benign clinical disorder has a risk of recurrence and therefore, for patients without reproductive desire, total hysterectomy with salpingooophorectomy and debulking can be performed as much as possible based on the risk assessment. Further hormonal suppression depends upon the degree of clearance given from DLP.

CONCLUSION

DPL is a rare benign clinical disorder which mimics disseminated peritoneal malignancies. The possibilities of DPL should be kept in mind while evaluating such cases when a history of uterine fibroids and prior uterine surgery is documented.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Rajeswari KSR, Nandhana V. Disseminated peritoneal leiomyomatosis-diagnostic dilemma in an acute presentation-a case report. Int J Reprod Contracept Obstet Gynecol 2023;12:2575-2577.