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Case Report

A case report on disseminated peritoneal leiomyomatosis

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ABSTRACT

Disseminated peritoneal leiomyomatosis (DPL) is a rare benign Smooth muscle tumor proliferating along the sub peritoneal and peritoneal surface. The pathogenesis of the disease remains unknown, but iatrogenic implantation, hormonal effects, heredity, and peritoneal mesenchymal stem cell metaplasia has been implied. Approximately 200 cases have been reported worldwide. Owing to the rarity of the disease and the lack of typical clinical symptoms and signs, the misdiagnosis rate is extremely high. We present a case of DPL intervened in our centre.

Keywords: Disseminated peritoneal leiomyomatosis, Benign smooth muscle tumour, Peritoneal mesenchymal stem cell metaplasia

INTRODUCTION

Disseminated peritoneal leiomyomatosis (DPL) is a rare benign smooth muscle tumor proliferating along the sub peritoneal and peritoneal surface.¹ The actual incidence of DPL might be underestimated, considering its asymptomatic nature.² The pathogenesis of the disease remains unknown, but iatrogenic implantation, hormonal effects, heredity, and peritoneal mesenchymal stem cell metaplasia has been implied. The condition was first described in 1952 by Wilson and Peale, later it was named by Taubert.³ We present a case of DPL intervened in our centre.

CASE REPORT

A 38-Year-old women with P1L1, has come with the complaints of abdominal discomfort and increased abdominal size since 1 month. she had regular cycles with history of laparoscopic myomectomy elsewhere 8 year back and also gives a history of morcellation during the surgery.

Clinical examination revealed a firm uterine mass of 20 weeks size. Per vaginal examination showed the uterus to be of 20-week size with restricted mobility. A firm mass, separate from the uterus was felt through bilateral fornix.

All laboratory investigations were within normal limits.



Figure 1: CT-scan of large heterogenous enhancing lesion in the right adnexa.

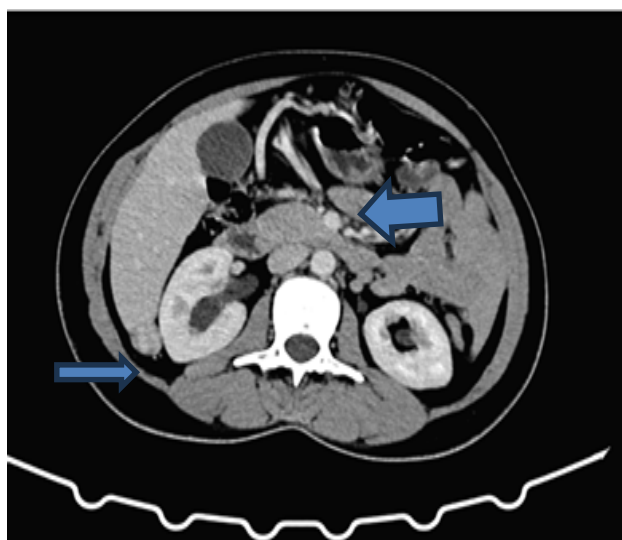


Figure 2: Small heterogenesis nodule seen along the Morison pouch and along the greater omentum.

Ultrasound abdomen and pelvis revealed a pedunculated sub serosal fibroid (18.3×9.4×13.3 cm) and hetero echoic lesion adjacent to distal end of abdominal aorta.

CT abdomen showed large heterogenous enhancing lesion in the right adnexa likely broad ligament fibroid and heterogenic lesion in mesentery along the surface of segment VI of liver and left subphrenic region likely DPL and mild hydroureteronephrosis due to pressure effect.

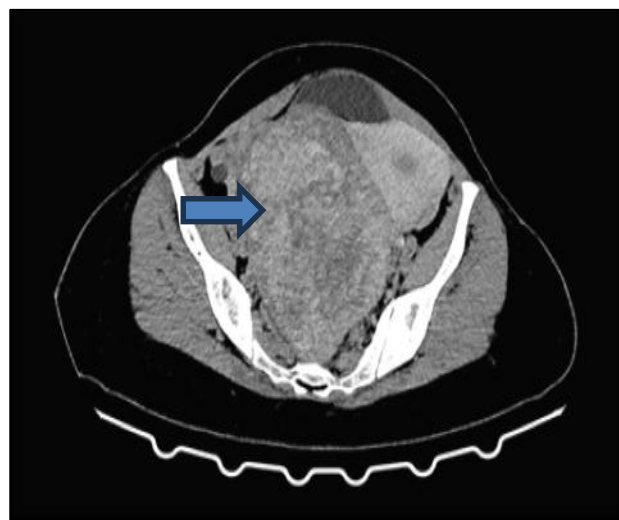


Figure 3: Large heterogenous mass displacing the uterus to the left

Post-contrast imaging also showed similar finding with parasitic fibroid in mesenteric plane of size (7.1×6.8 cm)

The imaging findings were suggestive of uterine leiomyoma with coexisting disseminated peritoneal leiomyomatosis. The patient underwent total abdominal

hysterectomy, bilateral salphingo-oophorectomy and removal of peritoneal leiomyomata.

Intraoperatively, uterus-14 weeks size with multiple subserosal and intramural fibroid, parasitic fibroid of size 15×12 cm is seen involving the seromuscular layer of sigmoid colon, fibroid of size 10×10 cm seen encased within greater omentum and ectopic fibroid of size 2×3 cm seen in the Morison pouch.

Histopathological examination of the specimens showed a tumour composed of interlacing bundles and whorls of benign spindle cells with focal areas of hyalinization, Multiple extrauterine lesions showed a whorled pattern separated from each other by vascularized connective tissue and no pleomorphism suggestive of benign leiomyomatous nodules with myxoid change in stroma.

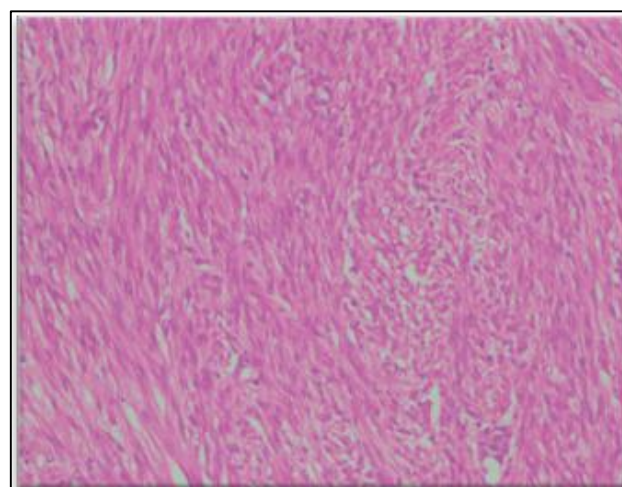


Figure 4: Tumor composed of interlacing fascicles of smooth muscle cells ×100 magnification H and E.

DISCUSSION

Leiomyoma are smooth muscle tumours that are common to the uterus, but uterine smooth muscle tumour with unusual growth pattern are rare and include 3 primary neoplasms: intravenous leiomyomatosis (IVL), benign metastasizing leiomyoma (BML) and disseminated peritoneal leiomyomatosis.⁴

The Etiology of DPL remains controversial and includes two main theories: Iatrogenic theory and hormone theory. The Iatrogenic theory hypothesizes that DPL is caused by the iatrogenic spread of myoma because of morcellation during myomectomy.⁵ DPL takes an average of 39-132 months to present after initial laparoscopic morcellation. It is estimated that the overall incidence of DPL after laparoscopic uncontained morcellation was 0.12-0.95%.⁶

During unconfined morcellation, small leiomyoma fragments and microscopic deposits, are easily dispersed and lost within the peritoneal cavity which in later life gets converted into DPL.⁷

DPL occurs mainly in the reproductive-aged females, and in some cases, they are found after use of oral contraceptives and hormonal replacement therapy, or during pregnancy and in the presence of an estrogen-secreting tumour.⁸

Most of the patients with DPL are asymptomatic. Others usually present with non-specific symptoms such as abnormal, heavy uterine bleeding and lower abdominal pain or discomfort. Less common presentations include increased frequency of micturition, mass per abdomen and symptoms of obstructive uropathy.

Imaging studies delineate the presence of intrauterine leiomyoma as well as the extent and location of peritoneal deposits. Ultrasonography and CT scan in patients with DPL show multiple, solid and complex soft tissue masses that are usually large and similar in morphology to uterine leiomyoma. MRI features include multiple masses with hypointense signal similar to that of skeletal and smooth muscles on T1 and T2 weighted images, which show variable post-contrast enhancement.⁹ Positron emission tomography is therefore a problem-solving tool in differentiating DPL from malignant peritoneal disease, which classically shows avid FDG uptake.¹⁰

Because of the rapid recovery and minimal trauma, laparotomy should be the first choice for surgical diagnosis and treatment. For patients with who desires fertility, focal resection is feasible, and aromatase inhibitors or gonadotropin-releasing hormone agonists are used post-operatively.¹¹ For patients without fertility requirements, resecting the entire uterus, bilateral appendages, and the abdominal mass is feasible, and resecting the greater omentum is performed if necessary.

CONCLUSION

DPL should be considered as a differential diagnosis of any intra-abdominal masses in patients with a history of myomectomy. Care should be taken during laparoscopic extraction of myomas with in-bag morcellation would be mandatory to avoid iatrogenic recurrence. Transvaginal morcellation through a posterior colpotomy incision has also been described following laparoscopic myomectomy and may be a valuable alternative to traditional morcellation methods.

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Ethical approval: Not required

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