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CASE REPORT: DISSEMINATED PERITONEAL LEIOMYOMATOSIS POST LAPARASCOPIC MYOMECTOMY

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ABSTRACT

Uterine fibroids affect women during the middle and later reproductive years. Recently, with advancement in laparoscopic techniques including the use of power morcellation, there has been an increase reported cases of extra uterine myomas. Disseminated peritoneal leiomyomatosis (DPL) or leiomyomatosis peritonealis disseminata is a rare condition, which is characterized by benign smooth muscle cells proliferation in the peritoneal cavity. This is a case of incidental intra operative DPL with synchronous recurrent uterine leiomyomas 6 years post uncontained laparoscopic myomectomy and morcellation. Histological of both uterine and extra uterine masses were consistent with benign leiomyomas.

INTRODUCTION

Uterine fibroids are benign monoclonal tumors of smooth muscle origin and the most common pelvic tumors in women(1). DPL is a rare and benign disease characterized by multiple smooth muscle nodules in the peritoneal surface or omentum, mesentery, colon, small intestine and ovary. The prevalence is estimated to be < 1 per 1,000,000 with only about 150 cases reported so far(2)(3). Postulated pathogenesis of DPL include

iatrogenic post laparoscopic uncontained morcellation, sub peritoneal mesenchymal stem cell metaplasia, hormonal and genetic factors.

Post myomectomy DPL is thought to arise from deposition of tissue fragments in peritoneum following power morcellation. Fragments can regrow and develop into parasitic fibroids. Incidence of parasitic fibroids post laparoscopic myomectomy with morcellation is estimated to be between 0.12% and 0.95%(4). Diagnosis is usually made at

surgery and confirmation is through histopathological and immunohistochemical staining.

Regionally this is the first case of DPL an incidental intra operative finding during open myomectomy for recurrence of symptomatic uterine fibroids 6 years post laparoscopic myomectomy.

CASE HISTORY

A 37-year-old female Para 0+1presented with secondary infertility for 13 years despite adequate coital frequency. She had recurrent symptomatic uterine fibroids; she underwent laparoscopic myomectomy with uncontained morcellation 6 years prior. Subsequent hysterosalpingiogram showed patent right fallopian tube and followed by three cycles of ovulation induction attempts. She however lost to follow up in the fertility clinic citing financial constraints. She presented 7 months prior to second surgery with dysmenorrhea but had normal regular menses. evacuation for an incomplete miscarriage was done 12 years prior. No family history of uterine fibroids reported. She was HIV negative with a normal pap smear.

She was in good general condition. Per abdomen she had 3 port laparoscopic scars and a firm, hard, non-tender pelvic mass corresponding to 24 weeks of gestation. Pelvic

examination revealed blood-stained foulsmelling discharge, the cervix was displaced to the left. Pelvic ultrasound showed multiple intramural fibroids the largest two measuring 10.3cm by 9.2cm and 7.4cm by 4.5cm and a sub serous fibroid measuring 7.9cm by 5.9cm. She was scheduled for open myomectomy.

Intra operative findings were of an enlarged deformed uterus corresponding to 24 weeks of gestation in size. There were three large myomas approximately 8 by 7cm each which occupied both the fundus and lower uterine segment. There were also multiple small sized fibroids averaging 3 by 2cm each exercised under diluted vasopressin. On exploration, there were two incidental benign looking masses; one on the sigmoid mesentery measuring about 5 by 3cm (figure 1) and the other attached mainly greater omentum and transverse mesocolon approximately 10 by 15 cm (figure 2) that were also excised with the help of a general surgeon. Two more incidental small sized benign looking masses (3 cm by 2 cm) were exercised from the anterolateral abdominal wall peritoneum. Other pelvic and gastrointestinal structures were grossly normal and there was no para-aortic or pelvic lymphadenopathy.

Histology for all masses showed smooth muscle fiber without atypia or necrosis. The masses were put in separate, well labelled containers for histopathology.

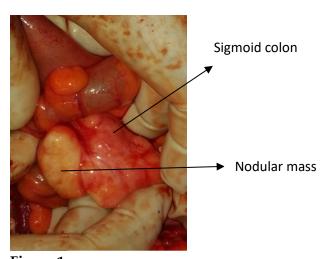


Figure 1: Firm nodular mass on sigmoid mesocolon

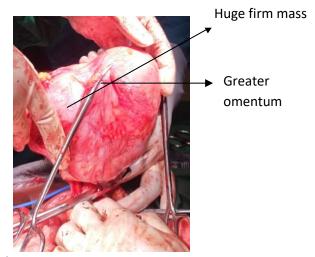


Figure 2: 10cm by 15 cm mass was being dissected from omentum.



Figure 3: Huge multiple uterine myomas

MICROSCOPY

Sections from the uterine masses showed a benign tumour composed of interlacing bundles of smooth muscle cells disposed in fascicles and whorling pattern in some areas. Hyalinized benign smooth muscles disposed in a fascicular pattern were seen in sections from the non uterine masses (omental, sigmoid mesentery and visceral peritoneal masses). Analogous to uterine leiomyomas, degenerative changes e.g. fibrosis, calcification, hyaline and cystic changes of soft tissue leiomyomas have been described (5).

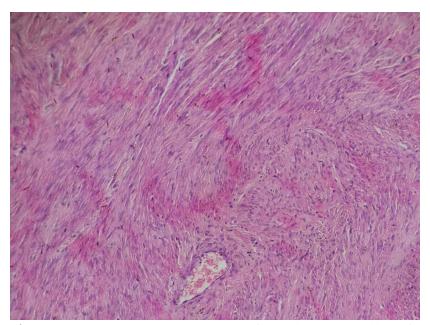


Figure 3: Benign tumor composed of interlacing bundles of smooth muscle cells disposed in fascicles.

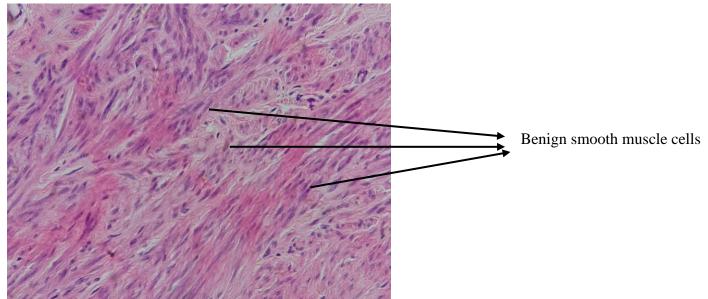


Figure 4: Smooth muscle cells with benign characteristics.

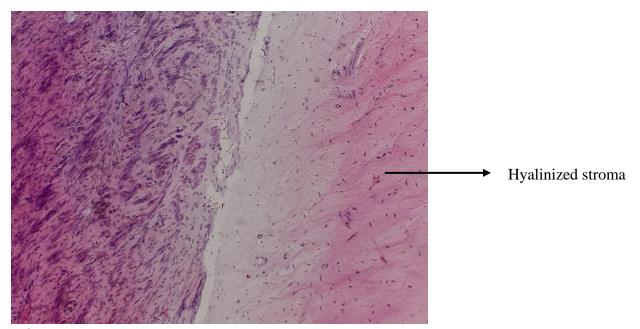


Figure 5: Stromal hyalinization within the tumour.

DISCUSSION

Disseminated peritoneal leiomyomatosis (DPL) is a rare entity characterized with multiple myoma nodules on the peritoneum. DPL presents a diagnostic challenge to surgeons and pathologists due to mimicry with disseminated intra abdominal malignancies, tuberculosis, peritoneal carcinomatosis or metastatic leiomyosarcoma. First described by Wilson and Peale in 1952, DPL is benign on histology although the risk of malignant degeneration of 10% has been reported(2)(3).

Progression of DPL is observed in states of increased estrogen such as pregnancy, combined hormonal pills, or ovarian estrogen secreting tumors suggesting the role of estrogen in its etiology. Progesterone and estrogen receptors demonstrated on the nodule smooth muscles in prior studies also support the theory of the role of steroid hormones pathogenesis of disseminated leiomyomatosis(6–8)

Ours is a case of DPL involving the peritoneal surface of the pelvic anterior abdominal wall, greater omentum, transverse and sigmoid mesocolons with synchronous recurrent uterine leiomyomas. The finding of multiple nodular masses can be supported by theory of iatrogenic seeding after the index laparoscopic myomectomy due to dissemination of the tumor cells along the laparoscopic tract. This safety concern especially for undiagnosed leiomyosarcoma informed the approval of a new tissue containment system(PneumoLiner) for morcellation in selected patients by the US Food and Drug administration in 2016(9). Our patient underwent uncontained laparoscopic myomectomy 6 years before the current laparotomy. She is currently scheduled for close long term follow up since her ovaries were left intact.

The extra uterine masses were incidental intraoperative findings although the patient had ultrasound evaluation which is valuable in aiding diagnosis. The lesions are often heterogeneous on CT attenuation, with enhancement that is similar to uterine leiomyomas and with low signal intensities on T2 weighted images and gradual and intense enhancement on magnetic resonance(10).

DPL cases are commonly asymptomatic. Common symptoms include abdominal distension, bloating, pain and nausea(2). Our patient was asymptomatic.

The definitive treatment of DPL is surgical resection of lesions. Other options include oophorectomy, use of GnRH analogues and aromatase inhibitors(2,6,8).

CONCLUSION

DPL although rare, should be considered in differential diagnosis of female abdominal and pelvic tumors/masses. Surgeons should counsel patients on the iatrogenic risk of DPL post myomectomy. Careful exploration, inspection, peritoneal washing and contained morcellation should be considered.

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