

Coexisting leiomyomata peritonealis disseminata and ovarian leiomyoma

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ABSTRACT

Leiomyoma is the most common tumor seen affecting women mostly of reproductive age groups. It is composed of smooth muscle cells that are positive for smooth muscle actin on immunohistochemistry. Leiomyomata peritonealis disseminata (LPD) is characterized by nodules that stud the peritoneum. It is seen in association with hormone-producing ovarian tumors and coexisting leiomyomas of the uterus. Association with mesenteric leiomyomas has been reported. Ovarian leiomyomas are very rare tumors of the ovary and coexisting LPD and ovarian leiomyomas has not been reported before. We present this rare case of LPD and ovarian leiomyoma.

Key Words: Leiomyomatosis peritonealis disseminata, mesentery, ovary, retroperitoneum

INTRODUCTION

Leiomyoma is the most common tumor in women. Myometrium of the uterine corpus is the most common site. Leiomyomas arise in the reproductive age group and are composed of interlacing bundles and fascicles of smooth muscle cells. They can also arise in the cervix, broad ligament, retroperitoneum, and mesentery. Leiomyomas with unusual growth patterns include benign metastasizing leiomyoma, retroperitoneal leiomyomas, parasitic leiomyomas, and LPD.^[1] Leiomyomatosis peritonealis disseminata is a very rare condition that is characterized by multiple nodules that stud the peritoneum and may also involve the mesentery. This condition arises usually a result of altered hormonal levels and occurs in pregnant women.^[2,3] We present a case of LPD in a postmenopausal woman involving the retroperitoneum, mesentery, and left ovary with a history of total hysterectomy 6 years back. To

the best of our knowledge, this is the first case of LPD with ovarian involvement reported in literature.

CASE REPORT

A 42-year-old multiparous woman presented with episodes of vague abdominal discomfort predominantly on the left side. Per abdominal examination revealed an ill-defined lump in the abdomen arising out of pelvis stretching beyond the umbilicus more on the left side. On pelvic examination, a pelvic mass that was of around 14 weeks gestational size, immobile, slightly tender, more on the left side was palpable. Her last childbirth was 19 years ago and she had undergone a total hysterectomy for a 20 cm fibroid arising from isthmus of the uterus 6 years back. The hematological and biochemical investigations including CA125 levels were within normal limits. Chest X-ray was normal.

Ultrasonography revealed mixed echogenicity mass in pelvis extending bilaterally, predominantly on left side,

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bilateral adnexae could not be visualized separately from the mass, the uterus was not seen as she had undergone a total hysterectomy. There was no free fluid in the pouch of Douglas [Figure 1a]. The upper abdomen was normal. Possibility of complex ovarian mass most likely cystadenoma of ovary was suggested and the patient was taken up for laparotomy. Peroperatively, a single nodule of 5 cm × 5 cm was also identified in the mesentery. The left ovary was enlarged and homogenous around 10 cm × 14 cm, posteriorly going into the retroperitoneum. Right ovary and tube were healthy looking. Tissue was taken for frozen sections from the retroperitoneum and ovary and possibility of a mesenchymal lesion was suggested. Subsequently, the nodules were excised and bilateral salphingoopherectomy was performed and specimen received in 10% formalin for histopathology. Grossly, the retroperitoneum, left ovary, and mesenteric nodules were gray-white, homogenous in appearance, the cut section showed a whorled gray-white appearance [Figure 1b]. The right ovary and bilateral tubes were unremarkable. Several tissue blocks were processed and routinely stained for light microscopy examination.

Histology revealed similar findings from the retroperitoneum, left ovary, and mesenteric masses; interlacing and intersecting fascicles of smooth muscle cells arranged in whorls and having an abundant eosinophilic fibrillar cytoplasm with focal myxoid and cystic change were appreciated [Figure 2a]. Areas of hyalinization were also present. There was no atypia or necrosis. Histomorphology was suggestive of ovarian leiomyoma and leiomyomatosis peritonealis disseminata (LPD). On Masson trichome stain, the smooth muscle cells stained red and the hyalinized areas were blue [Figure 2b]. The smooth muscle cells were positive for smooth muscle actin (SMA) on immunohistochemistry [Figure 2c]. The smooth muscle cells were also positive for estrogen receptor (ER) and progesterone receptor (PR) which further supported the diagnosis.

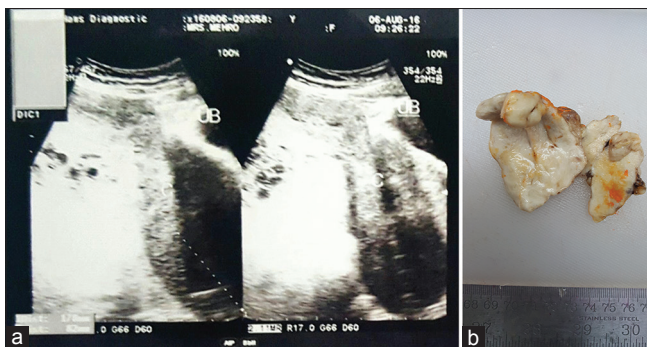


Figure 1: (a) Ultrasound showing a mixed echogenicity mass in the pelvis. (b) Cut section of the retroperitoneal nodule showing gray-white whorled appearance

DISCUSSION

Leiomyoma is the most common tumor of the female genital tract, seen mostly in women of reproductive age group. It arises from the smooth muscle of the myometrium and is classified as submucosal, intramural, or subserosal. The cervix, broad ligament, mesentery, and retroperitoneum are the other sites of its origin. LPD also called diffuse peritoneal leiomyomatosis a very rare disease that is characterized by multiple nodules studding the peritoneum.^[2,3] The first case was reported by Willson and Peale in 1952. Wilson *et al.* coined the term LPD in 1965.^[4] Abnormal hormonal environment seems to play a role and this explains why this condition is seen in women of reproductive age group with many of them discovered during pregnancy,^[2-4] with estrogen stimulating the mesenchymal cells to differentiate into fibroblasts, myofibroblasts, and smooth muscle.^[5,6] Most of the patients also have concomitant uterine leiomyomas. It may arise in the setting of estrogen-secreting ovarian tumors.^[4] Ovarian leiomyoma is an extremely rare tumor of the ovary with only sixty cases reported in literature.^[5-7] Association of LPD with ovarian leiomyoma has not been reported in literature. The present case was a 42-year-old lady who had coexisting ovarian leiomyoma with LPD, with total hysterectomy for uterine leiomyomas done 6 years back. Exposure to exogenous hormones, hormone-secreting ovarian tumors, development after laparoscopic morcellation of myomas are the other factors that are described in the development of LPD.^[5,6] The presence of ER and PR has been described in LPD and spontaneous regression noted after menopause. The retroperitoneal and mesenteric nodules and left ovary in our case showed whorls and fascicles of smooth muscle cells with cystic and myxoid degeneration.

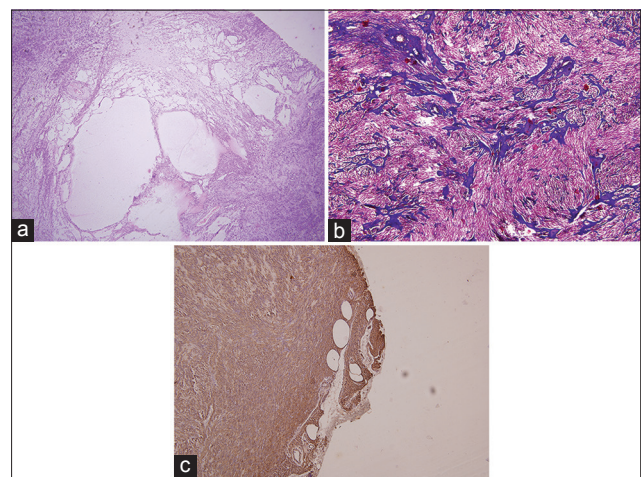


Figure 2: (a) Photomicrograph showing whorls and fascicles of smooth muscle cells along with myxoid and cystic change (H and E, ×100). (b) Photomicrograph showing red and blue taken up by smooth muscle and hyalinized areas, respectively (Massons trichome, ×400). (c) Photomicrograph showing smooth muscle actin positivity in smooth muscle cells (IHC, ×400)

There was positivity for ER and PR. No recurrence was reported in the patient after 2 months of surgery.

Possibilities of malignant ovarian tumor, fibromas, thecomas, peritoneal carcinomatosis, leiomyosarcoma, and gastrointestinal stromal tumor considered.^[5-7] There was no cytological or nuclear atypia, and the ovarian tissue was completely replaced by smooth muscle proliferation; hence, malignant ovarian tumor was excluded. Positivity for SMA excluded thecomas and fibromas. The upper and gastrointestinal tract was normal on scans; hence, possibility of metastasis or peritoneal carcinomatosis was nulled. There was no necrosis or mitosis, thus leiomyosarcoma was excluded. To conclude, malignant transformations in LPD though rare have been reported; hence, follow-up of patients is necessary. LPD with coexisting ovarian leiomyoma can simulate a malignancy and result in extensive dissections resulting in morbidity and mortality.

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Conflicts of interest

There are no conflicts of interest.

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