CASE REPORT

Leiomyomatosis peritonealis disseminata in a Nigerian woman

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ABSTRACT

Leiomyomatosis peritonealis disseminata (LPD) is a rare condition. A 48-year-old multiparous woman was referred because of an incidental ultrasound finding suggestive of LPD. She had a 6-year past history of use of combined oral contraceptive pills. LPD was also suspected at laparotomy and confirmed by histology. She had total abdominal hysterectomy, bilateral salpingo ophorectomy and infracolic omentectomy. Patient was being followed-up. LSD is a rare gynecological condition which can pose a diagnostic challenge. Removal of estrogen sources as was done for the patient is the mainstay of treatment. Patient follow-up is very important because of the risk of malignant transformation.

Key words: Laparatomy, leiomyoma, leiomyomatosis peritonealis disseminata, Nigeria, ophorectomy

INTRODUCTION

Leiomyomatosis peritonealis disseminata (LPD) is a condition characterized by numerous leiomyomas throughout the peritoneal cavity which appears grossly malignant but histologically benign.[1] It is a very rare gynecological condition.[2]

CASE REPORT

The patient was a 48-year-old woman who was referred to the gynecology clinic of a teaching hospital in Nigeria because of an ultrasound report suggestive of LPD. She was para 3 + 0 with 3 living children. She had noticed a slowly progressive abdominal swelling of about one year duration which made her to carry out the abdominal ultrasound. She was first seen at the hospital in June, 2008, and had no other symptoms. Her menarche was at the age of seventeen years and she had a regular 28-day menstrual cycle with a 5-day normal menstrual flow. She had not noticed any weight loss. She had used combined oral contraceptives (COC) for about 6 years for child spacing. Her last confinement was in 1993. There was no family history of fibroid, genital or breast cancers.

There was no abnormality on general examination. Her abdomen was full, nontender, and moved with respiration. The liver, spleen and kidneys were not palpable. There was a firm, nodular and nontender suprapubic mass of about 20 weeks' gestation size. There was no shifting dullness. On vaginal examination, there was no vaginal bleeding. The cervix appeared healthy and moved with the abdominal mass. The lateral and posterior vaginal fornices were occupied by firm, nontender, and mobile masses. There were no significant findings on examination of the respiratory and cardiovascular systems. A working diagnosis of uterine fibroid coexisting with LPD was made to rule out ovarian malignancy. Results of her full blood count, chest radiogram, renal, and liver function tests were normal.

The ultrasound report showed "Bulky uterus measuring 18×7.5 cm in anteroposterior dimension. It harbours multiple heteroechoic well circumscribed masses—leiomyomata. A subserosal mass bulges into the posterior wall of the bladder and measures 2.8×3.9 cm. Multiple similar masses are also seen scattered in the abdominal cavity. A right hypochondrial/subhepatic mass measures 10.5×8.0 cm. An epigastric mass: 3.6 cm×3.8 cm. Left hypochondrial mass: 9.9×7.2 cm. Three left flank masses subjacent to the spleen are seen measuring 5.0×3.4 cm, 4.8×3.1 cm, 2.5×3.0 cm. The spleen was not well visualized. The liver, gall bladder, and both kidneys appear sonographically normal." She consented to exploratory laparotomy.

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At laparotomy, there was no ascitis. The uterus was enlarged with fibroids and there were innumerable grey-white, firm nodular seedlings of varying sizes attached to the adnexa and the greater omentum [Figures 1 and 2]. She had a total abdominal hysterectomy, bilateral salpingo-ophorectomy, and infracolic omentectomy. Histology of surgical specimen showed "benign smooth muscle lesions composed of bundles of mature smooth muscle cells devoid of mitosis or atypia separated by thin fibromuscular connective tissue septae"; and confirmed the diagnosis of LPD.

She was being followed up routinely and there had not been any physical or radiologic feature of the disease progression.

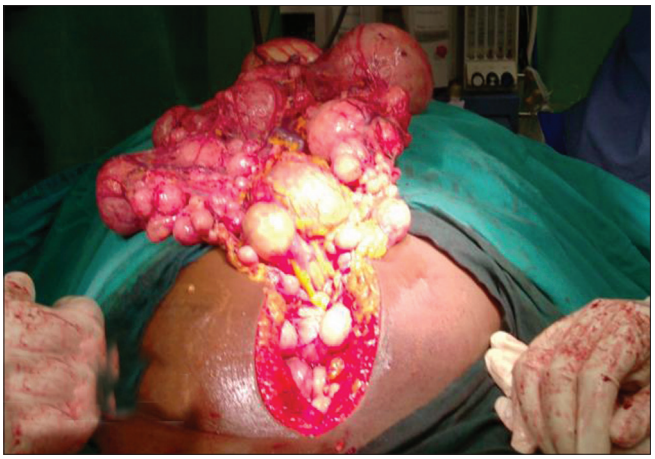

Figure 1: Leiomyomatosis peritonealis disseminata

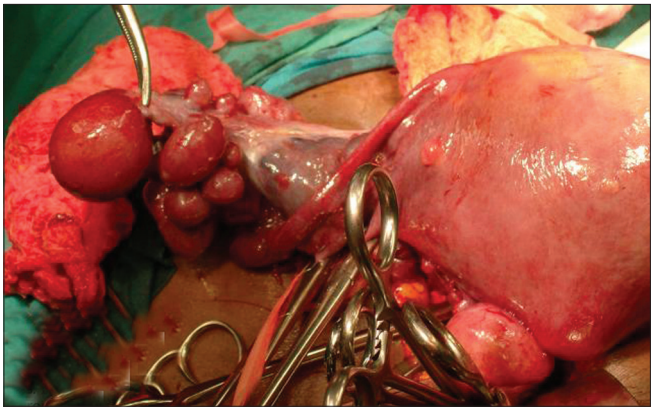

Figure 2: Leiomyomatosis peritonealis disseminata and uterine fibroid

DISCUSSION

LPD or diffuse peritoneal leiomyomatosis is a benign disorder, characterized by the development of numerous myomas throughout the peritoneal cavity.[3] As in this patient, it is common in reproductive age women,[4] but can also occur in postmenopausal women,[3] and men.[4] The etiology of LPD is unknown, but animal study suggests that it may be related to the metaplasia of submesothelial, pluripotent mesenchymal cells after prolonged exposure to high level of estrogen with or without progesterone.[5] This hormonal pathophysiology is supported by a recent report which showed a spontaneous and total regression of the tumor in a pregnant woman after delivery.[6] The prolonged exposure of the patient to COC might be related to the disorder in this case, since the condition is associated with high levels of exogenous or endogenous female gonadal steroids.[7] Despite the high prevalence of uterine fibroid and high fertility rate in Nigeria, only one case of LPD has been reported.[8] Though reporting bias cannot be ruled-out completely, it is possible that the condition may be more related to exogenous hormones considering the fact that the use of COC is very low in Nigeria. Also, as was evident in the patient, LPDs are usually asymptomatic and often coexist with uterine fibroid.[7] Furthermore, the condition may mimic malignancies hence posing serious diagnostic challenge.[9] However, the absence of weight loss and ascitis as well as the sparing of the liver that characterized this case were important negatives against peritoneal carcinomatosis. Radiological investigations (ultrasound, CT Scan, MRI) help in diagnosis but may not be able to differentiate other differentials such as peritoneal carcinomatosis, LPD with sarcomatous change, lymphadenopathy, multiple pedunculated leiomyomas. Definitive diagnosis of LPD relies on histological examination of tissue specimen as was done in this case.

The main stay of treatment for LPD involves the removal of source(s) of estrogen/progesterone which could be surgical, medical, or termination of pregnancy as the case may be. Nevertheless, treatment should be guided by the patient's age, symptomatology, hormonal and reproductive status.[10] The patient was perimenopausal and had no future reproductive wishes; therefore, the option of surgical castration was appropriate for her. Nevertheless, the unavailability of intraoperative frozen section at the hospital which should direct the extent of surgeries in cases of suspicious intra-operative findings would have made uterine/ovarian saving surgery impractical in this case because of the risk of leaving behind palpable malignant tissue. Different drugs such as gonadotropin releasing hormone agonist, megestrol acetate, danazol, and raloxifene have been used for LPD treatment in some cases but with poor results.[1,10] However, aromatase inhibitor—anastrozole may be effective for the control of tumor growth and its symptoms.[1]

Though malignant transformation is very rare, it has been noted up to 8 years after diagnosis of the condition.[10] Because of this risk of malignant transformation and that of disease recurrence, there was the need for regular clinical and radiological follow-up of the patient.

In conclusion, LSD is very rare in Nigeria. Removal of hormone source(s) as was done for the patient remains

the mainstay of treatment. Patient follow-up is important because of the recurrence and carcinogenic potentials of the tumor.

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