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Case Report

Leiomyomatosis Peritonealis Disseminata: Case Report

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ABSTRACT

Background: Leiomyomatosis Peritonealis Disseminata (LPD) is a very rare smooth muscle tumour characterized by dissemination of multiple smooth muscle - like nodules throughout the omental and peritoneal surfaces. Before now, only two cases seem to have been reported in Nigeria. It may mimic disseminated malignant peritoneal metastatic nodules. The aetiology of LPD include hormonal genetic and iatrogenic theories. It's link to estrogen / progesterone stimulation is, however, strong. **Setting:** Imo State University Teaching Hospital, Orlu, Imo State, Nigeria. **Case report:** A case of a 49-year-old para2 woman who presented with severe menorrhagia and dysmenorrhoea. A diagnosis of uterine leiomyoma was made for which she had Total Abdominal Hysterectomy plus Bilateral Salpingo-oophorectomy (TAH+BSO). Incidentally, during the surgery, multiple widespread leiomyoma-like nodules were noted in the peritoneal and omental surfaces. These were meticulously excised and histology report confirmed LPD. Curiously, this woman developed Non-Insulin Dependent Diabetes Mellitus about one month after surgery raising a question whether Diabetes mellitus is a probable complication of LPD. **Conclusion:** The possibility of LPD coexisting with a fibroid-riddled uterus should be borne in mind whenever surgery is being carried out for uterine leiomyoma.

Keywords: leiomyomatosis peritonealis disseminata coexisting with uterine leiomyoma, Orlu, South East, Nigeria.

Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a very rare benign smooth muscle tumour characterized by dissemination of multiple smooth muscle-like nodules throughout the omental and peritoneal surfaces. The first case of LPD was reported in 1952 by Wilson & Peale and they suspected it to be due an abnormal response to

ovarian hormonal stimulation, be it normal or elevated.[1] However, this condition was first clearly delineated and named by Taubert et al in 1965.[2]

A case is reported here in Orlu, South East Nigeria, probably the third case reported in Nigeria after the two previous cases reported by Dim et al[3] and Awolola et al,[4] discovered during hysterectomy for leiomyoma.

Case Report

A 49 year old para 2 presented in our institution in April 2018 with two-year history of recurrent heavy menstrual blood flow associated with dysmenorrhoea. She was mildly pale when examined and there was an abdominal mass equivalent to 20 weeks gestation in size. Her packed cell volume (PCV) was 24% and ultrasound scan reported a fibroid-riddled uterus containing subserous, intramural and submucous fibroid nodules. No other intra-abdominal masses were noted. She was counseled for total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH+BSO) which she consented to. She, however, did not turn up until a month later with severe anaemia (PCV was 12%) following severe heavy menstrual blood loss. TAH+BSO was promptly arranged. She was transfused with three units of packed red cells pre-operatively.

During the surgery a 20-week size uterus riddled with fibroid nodules was found. In addition, several fibroid-like nodules were seen adherent to intestinal loops, omentum and the bladder wall. (Figure 1-3)

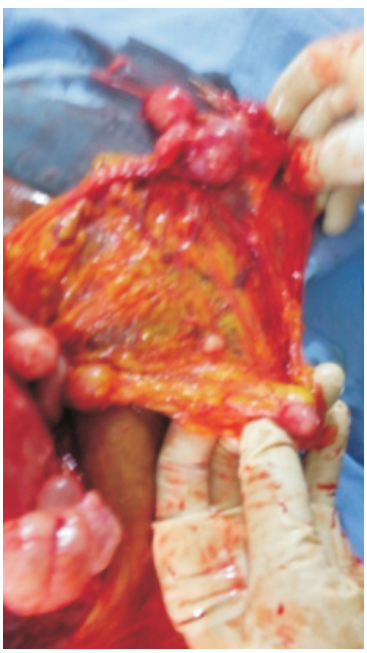

Figure 1
Leiomyomatous nodules (LPD) on the omentum.

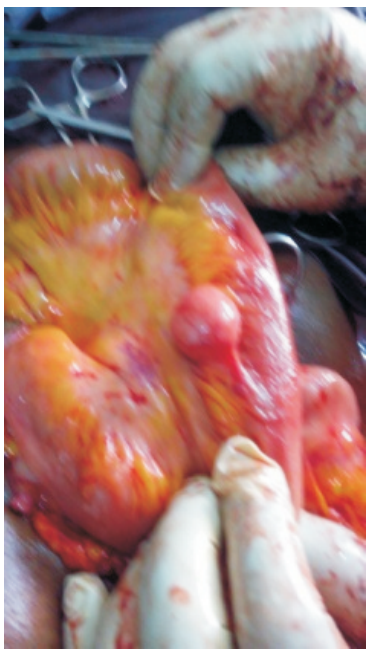

Figure 2
Leiomyomatous nodules (LPD) on the bowel Mesentery

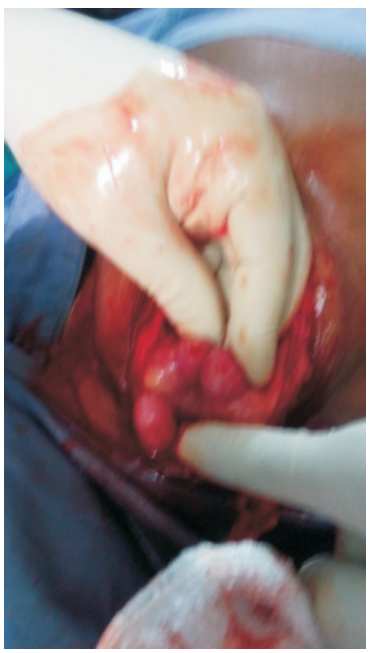

Figure 3
Leiomyomatous nodules (LPD) on the peritoneal covering of the bladder wall.

TAH+BSO was carried out. The fibroid like nodules (ranging in size from 1cm to 5cm) adherent to the bowel loops, the omentum and the bladder wall were carefully excised and meticulous haemostasis secured. She had two pints of whole blood intra-operatively. She was discharged home on post-operative day 5 in stable condition with a post-operative PCV of 24%.

Curiously, a month after she was discharged from the hospital, during her follow-up visit, she presented with polyuria and polydipsia which started few days prior to her presentation. She was confirmed to have developed Diabetes Mellitus (DM) with a fasting blood sugar of 13.6mmol/L. There was no history of DM prior to surgery and investigations carried out before the surgery revealed no sign of DM. She was managed with oral hypoglycemic agent and dietary advice and the DM was controlled. She was then referred to an endocrinologist for further management of DM. Histology report of the intra-abdominal fibroid-like nodules was "extra uterine masses with interlacing fascicles of smooth muscle cells arranged in whorls. No mitotic figures or nuclear pleomorphism suggestive of malignancy were noted" confirmed that they were leiomyomatous tissue, just like the uterine masses histology also revealed.

Discussion

LPD is most common in women in their reproductive years with 50% of the cases occurring in black women.[5] This case is another black woman and she was also in her reproductive years although towards the end of reproductive years. Rarely though, LPD has been reported in post menopausal women.[6] A case was even reported in a man, albeit, with malignant change.[7]

The association of this LPD case and fibroid-riddled uterus supports the theory that LPD is associated with increased sensitivity to estrogens1. Spontaneous regression of LPD in those cases associated with pregnancy or an estrogen producing tumour, after estrogen levels have normalized, supports this fact. Some workers have even experimentally induced extra-genital leiomyomas in guinea-pigs by exposing them to estrogen & progesterone.[8] Apart from this possible hormonal cause of LPD, other possible causes could be metaplasia of sub peritoneal mesenchymal stem cells, genetic, or morcellation of myoma during laparoscopic surgery with resultant residual fibroid tissues being left behind in the peritoneal cavity.[9]

The majority of patients with LPD are asymptomatic. Those that are associated with uterine leiomyomas may, however, present with common symptoms associated with uterine leiomyomas such as menorrhagia and dysmenorrhoea as seen in this case. When LPDs occur alone, symptoms, if any, are usually non-specific and they include abdominal pain and discomfort, bleeding from the rectum or the vagina, abdominal distention or abdominal masses which may lead to intestinal obstruction.[10]

Pre-operative diagnosis using imaging procedures such as abdominal ultra sound scan (USS), CT or MRI, is difficult probably because of low index of suspicion. The pre operative ultrasound scan done for this patient could not identify the disseminated leiomyomatous nodules in the peritoneal cavity. The only probable case diagnosed pre operatively by USS was reported by Singh G et al in 2002.[5] Diagnosis, as in this case, must be confirmed through histologic studies of the specimens. The histologic findings in this case were "extra-uterine masses with interlacing fascicles of smooth muscle cells arranged in whorls. No mitotic figures or nuclear pleomorphism suggestive of malignancy were noted". These findings were similar to the histology study findings of the uterine specimen.

The major differential diagnosis of LPD are peritoneal carcinomatosis, peritoneal metastic lesions and benign metastasizing leiomyoma (BML). Histologic studies will help to exclude the former two but histologic differentiation from BML is difficult. However, BML is considered as a smooth muscle mass in a solid organ11. While BML, when seen in the abdomen or the pelvis, typically is detected close to the round ligament or the iliac veins.[12] LPD is seen widespread in the peritoneal cavity involving the peritoneal & omental surfaces

of the abdominal cavity, the small & large intestines, the mesentery and the retroperitoneum.[13]

Although LPD is a rare benign tumour with a good prognosis, degeneration of LPD into malignancy has been reported in the literature.[14] Also, simultaneous identification of LPD and leiomyosarcoma has been reported, although it is not clear if the malignancy occurred due to transformation of the LPD tumour. It may therefore be necessary to institute guidelines for follow-up in patients with LPD, even after treatment especially in those cases considered to have high risk of malignant degeneration such as those with no prior exposure to estrogens, those with no previous history of leiomyoma and those with negative progesterone and estrogen receptors in the benign nodules.[15] Such high-risk cases may benefit from regular abdominal CT or MRI monitoring although care should be taken with too frequent CT monitoring as frequent exposure to CT radiation could predispose to other malignancies.

Finally, concerning treatment of LPD, recently, determination of the therapy according to the patient's age, comorbidities and severity of symptoms of LPD has been proposed.[12] Generally, for women with reproductive desire, a conservative therapeutic approach is preferred. More aggressive surgical treatment is recommended in cases of high risk of malignant degeneration.

Conclusion

LPD is a very rare peritoneal disseminating benign tumour which may be coexisting with uterine leiomyoma or occurring alone. This reported case is probably the third case reported in Nigeria.

Since uterine leiomyoma is very common in Nigeria, the possibility of LPD should be looked out for especially in cases where the uterus is riddled with leiomyomatous nodules.

Because LPD may mimic malignant peritoneal metastatic nodules, the presumed LPD nodules should be subjected to histologic studies so that appropriate treatment can be undertaken.

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