



A Rare Case of Leiomyomatosis Peritonealis Disseminata

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/IJMPCR/2023/v16i4341

Open Peer Review History:

This journal follows the Advanced Open Peer Review policy. Identity of the Reviewers, Editor(s) and additional Reviewers, peer review comments, different versions of the manuscript, comments of the editors, etc are available here: <https://www.sdiarticle5.com/review-history/103385>

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ABSTRACT

Leiomyomatosis Peritonealis Disseminata (LPD), also known as disseminated peritoneal leiomyomatosis is an extremely rare disease in women and men where there is a rapid increase in peritoneal and subperitoneal nodules mainly of smooth muscle cells. It is a benign condition, however, its diagnosis can mimic or degenerate into peritoneal leiomyosarcoma, making the diagnosis tricky. In addition, clinical manifestations can be very nonspecific.

We discuss the case of a 38-year-old nulliparous patient who presented to the clinic with a 2-year history of lower abdominal swelling and a 5-month history of severe menorrhagia. Intraoperative findings showed an enlarged uterus with subserosal fibroids and several deposits on the serosa and intestines. The patient had Total hysterectomy and bilateral-salpingoophorectomy. Because malignant transformation has been reported to potentially occur almost 10 years from initial diagnosis, patients should be monitored by repeat ultrasound scans.

Keywords: *Leiomyomatosis peritonealis disseminata; subserosal fibroids; malignant transformation; estrogen.*

1. INTRODUCTION

Leiomyomatosis Peritonealis Disseminata (LPD), also known as disseminated peritoneal leiomyomatosis is an extremely rare disease in women and men [1,2,3]. Patients with this condition have multiple leiomyomas in the peritoneal cavity which appear malignant but in reality, are benign [4,5]. Female patients are usually in their reproductive age, however there have been reports of this condition after menopause [1,6]. Some cases are discovered incidentally, during pelvic surgery, cesarean section or at autopsy [4,6]. Medical literature also suggests that prolonged estrogen exposure may be responsible and patients who have been pregnant, are pregnant, on oral contraceptives or hormone replacement are at risk of LPD [2,5]. Based on our search in the medical literature, there were only five documented cases of this condition in Nigeria [1,2,3,5,6]. Because many cases are unlikely to be reported, it is possible that the reported incidence for Nigeria is inaccurate.

2. CASE PRESENTATION

This is the case of a 38-year-old nulliparous lady who presented to the clinic with a 2-year history of lower abdominal swelling and a 5-month history of severe menorrhagia. According to her gynecological history, her menses lasted 10 days compared to her previous menses that lasted 5 days. Additional history included weakness, dizziness and shortness of breath on mild exertion.

Positive findings on general examination included- mild respiratory distress [28 cycles/min] and pallor. On examination of her abdomen, there was a 22-week lower abdominal mass described as firm, non-tender, and nodular, arising from the pelvic region.

On Vaginal exam, there was a soaked sanitary pad and the vulvar was smeared with blood. Speculum exam revealed blood from the uterine cavity with a healthy looking ectocervix. Bimanual exam showed a bulky uterus that moved away from the examining finger with no adnexal tenderness.

Investigations showed that hemoglobin was 6.5gm/dl while her electrolytes and urea as well as liver function tests were all normal.

Abdominal ultrasound revealed a very bulky non-gravid uterus with heterogeneous myometrial echo pattern. Endometrial thickness was not appreciated because of multiple myometrial masses. There were multiple masses seen mostly micro-myoma with the highest in size measuring 70 x 58cm [posterior submucous], 56 x 40cm [anterior submucous, 40 x 34cm [endometrial] & other micro-myometrial masses.

An impression of multiple myometrial masses was made. The patient was counselled on the diagnosis and treatment [myomectomy, possible subtotal hysterectomy]. She was admitted and worked up for surgery. She received 3 units of blood before the surgery.



Fig. 1. Morphology of Leiomyomatosis Peritonealis Disseminata (LPD)

Intraoperative findings showed an enlarged uterus with subserosal fibroids and deposits on the serosa and intestines. There were no areas of cleavage between the multiple fibroid nodules involving the whole uterine muscles and cavity. In addition, there were beaded fallopian tubes. Total hysterectomy and bilateral-salpingoophorectomy were performed.

The uterus and the ovaries were sent for histology with features suggestive of Leiomyomatosis Peritonealis Disseminata (LPD).

3. DISCUSSION

This uncommonly documented condition is common in reproductive aged women. It was first reported by Wilson and Peale as multiple peritoneal leiomyomas and the term Leiomyomatosis Peritonealis Disseminata (LPD) was established in 1965 [3,7,8].

Medical literature suggests two main factors that are responsible for LPD. Exposure to female sex hormones (estrogen and progesterone) in premenopausal women has been implicated i.e. estrogen and progesterone receptors. Iatrogenic etiology has also been implicated i.e., following laparoscopic myomectomy with morcellation considering that the procedure might result in residual myoma tissues. This second factor cannot explain our patient's case.

At the time of writing this report, only five cases in Nigeria were identified in the medical literature [1-5]. While this condition can mimic a malignant condition, it is a benign disorder where the patients have numerous myomas in the peritoneal cavity [2-4]. Some patients may have

coexisting fibroids. Our patient is of reproductive age; this condition is not common in postmenopausal women but it has been reported. It is important to note that no definite cause has been identified in the literature. As previously mentioned, medical literature implicates long term exposure to estrogen plus or minus progesterone. Pregnancy, hormone replacement therapy and oral contraceptive pills belong to this category of etiological factor for which our patient may belong to [4-6,9]. Many patients do not have symptoms, however, for those who have symptoms, they present with menorrhagia. Others have abdominal pain, abdominal swelling and/ demonstrable abdominal masses [1, 2, 10]. Investigating this condition includes imaging such as ultrasound scan or CT scan. Definitive diagnosis is made by histology. Treatment is typically surgery and is dependent on hormones, reproductive status, age and the symptoms. Our patient had Total hysterectomy and bilateral-salpingoophorectomy. Because malignant transformation has been reported to potentially occur almost 10 years from initial diagnosis, patients should be monitored by repeat ultrasound scans [2].

4. CONCLUSION

LPD is an uncommon and benign disease which can be confused for malignancy in the peritoneum. This condition should be suspected in a patient exposed to estrogen and/progesterone, or a patient with a history of myomectomy with morcellation who presents with abdominal mass. Even though malignant transformation has been rarely reported, it is a possibility. Patients with malignant transformation will possibly include: postmenopausal patients,

patients without a history of myomectomy with morcellation and male patients. Differentials for this condition include: disseminated leiomyosarcoma, peritoneal carcinomatosis and tumors of the gastrointestinal system. Treatment depends on different factors.

CONSENT

Per international standard or university standard, patient(s) written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

Per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Ezeome, Emmanuel R., Francesca Mannini, and Bankole D. Olusina. Progressive leiomyomatosis Peritonealis Disseminata (LPD): A case report and review of literature. *Tropical Journal of Obstetrics and Gynaecology*. 2005;22:197-199.
2. Ndububa Victor. Leiomyomatosis peritonealis disseminata: Case report. *Tropical Journal of Obstetrics and Gynaecology*. 2020;37(3):575-579.
3. Lanre Awosusi, Adegoke Oluwafadekemi, Nwanji Dupe, Oni Fola. Disseminated peritoneal leiomyomatosis: A case report of an incidental finding during an emergency caesarean section and a review of the literature. *Asian Journal of Medicine and Health*. 2021;19(3):37-42.
4. Awolola OO, Ogbuokiri CM. Leiomyomatosis peritonealis disseminata an incidental finding during an emergency caesarean section in a private health care facility in Lagos, Nigeria. *Tropical Journal of Obstetrics and Gynaecology*. 2015; 32(1):155-158.
5. Dim Cyril C, Sunday P Akogu, Hyginus U Ezegwui, Daniel B Olusina. Leiomyomatosis peritonealis disseminata in a Nigerian woman. *Nigerian Medical Journal: Journal of the Nigeria Medical Association*. 2012;53(3):172.
6. Kim H, Cheong H. Leiomyomatosis Peritonealis Disseminata: An Incidental Finding at Autopsy and Review of Literature. *The American Journal of Forensic Medicine and Pathology*. 2023; 44(2):e10-2.
7. Izi Z, Outznit M, Cherraqi A, Tbouda M, Billah NM, Nassar I. Disseminated peritoneal leiomyomatosis: A case report. *Radiology Case Reports*. 2023;18(6):2237-40.
8. Diaz ES, Pereira GJ, Pareja R, Rodriguez OG, Ortíz CA, Duran CP. Recurrent leiomyomatosis peritonealis disseminata. *International Journal of Gynecologic Cancer*. 2023;33(5).
9. Bagri N, Aggarwal A, Misra R. Disseminated Peritoneal Leiomyomatosis with Sarcomatous Transformation: A Rare Case Report and Diagnostic Enigma. *Indian Journal of Gynecologic Oncology*. 2023;21(2):1-3.
10. Chang CC, Lin YH, Su KM, Yu MH. Leiomyomatosis peritonealis disseminata resembling intra-abdominal malignancy. *Journal of Medical Sciences*. 2023;43(2): 87.

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Peer-review history:
The peer review history for this paper can be accessed here:
<https://www.sdiarticle5.com/review-history/103385>