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Leiomyomatosis Peritonealis Disseminata - Repeat Abdominal Myomectomy With Hormonal Therapy: A Case Report From South-West Nigeria

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Case Report

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Abstract

Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a rare, typically benign condition characterized by multiple smooth muscle nodules proliferating along visceral and parietal peritoneal and abdominopelvic surfaces. Just a little over 200 cases have been reported worldwide since the condition was first described in 1952. This case highlights the possibility of finding LPD following previous abdominal myomectomy.

Case presentation

A 41-year old nullipara with recurrent fibroid with menorrhagia, presented to the gynaecology clinic with huge abdominal mass, for repeat abdominal myomectomy. Preoperative assessment was done with physical examination and investigations. Following abdominal myomectomy, incidental appendectomy and random excision biopsy of widespread tiny nodules on the gut mesentry and mesoappendix; histopathological assessment revealed disseminated leiomyoma nodules with an isolated focus of endometriosis.

Conclusion

LPD is a distant rarity, however, a high index of suspicion is required when a woman with risk factors for the condition is having myomectomy (especially a repeat myomectomy). The excision biopsy of peritoneal nodules should be subjected to histopathologic assessment to clinch the diagnosis of LPD.

INTRODUCTION

Leiomyomatosis peritonealis disseminata is a rare benign condition.[1] It is characterized by numerous small smooth muscle nodules (like leiomyoma nodules) growing on the visceral and parietal peritoneum of the abdomen and pelvis. It can involve the leaves of the broad ligament, including the mesosalpinges, mesoovarii; the mesentry of the gut including the mesocolon, mesoappendix, and the omentum.[2] It may often appear like malignant metastasis intra-operatively, or milliary tuberculosis of the abdomen, because of the widespread deposits of small sometimes very tiny nodules as was seen in the index patient. So far only about two hundred cases have been recorded all over the world.[3] In Africa there is very scanty documentation of this condition. Though this may be due to failure to report such cases or under-reporting, and limited access to required investigations both pre-operative and post-operative.[3] It is usually found in women of reproductive age, and very few cases have been reported in Nigeria.[4–7] Although 2 cases reported in South-East and North-central Nigeria were in a multiparous peri-menopausal and nulliparous premenopausal woman respectively.[8,9] Two other previous cases reported prior to these were an incidental finding at emergency caesarean section, and in a perimenopausal multiparous woman respectively.[10,11] These are apparently the only four cases that have been reported in Nigeria prior to this index case.

Risk factors for this condition include the female reproductive hormones (oestrogen and progesterone), growth of multipotent stem cells in the abdomen, and previous myomectomy especially laparoscopic myomectomy in which morcellation was employed. Patients may have no symptoms, and may be discovered incidentally during abdominal or pelvic surgeries like caesarean section or abdominal myomectomy as was the case with our index patient who was having a repeat abdominal myomectomy from recurrent huge uterine fibroid. For symptomatic patients, the symptom may be mild to severe abdominal pain with or without abdominal swelling, and pelvic pressure. The nodules may vary in size, between 0.1 to up to 6cm in widest diameter.[5] The index patient had widespread tiny peritoneal nodules with largest diameter about 0.1 to 0.5cm. Due to the extreme rarity of the condition, it eludes suspicion preoperatively. Investigations that may be required to make a diagnosis include radiological investigations like ultrasonography (transvaginal and/or transabdominal), computerized tomographic (CT) scan, magnetic resonance imaging (MRI); post-operative tissue biopsy as in the index case, or ultrasound-guided biopsy; this will confirm the cells are benign smooth muscle cells, and rule out the typical caseating lesions of milliary tuberculosis (diagnosis of which will be accompanied by prominent clinical features, with laboratory tests like the gene X-pert test).[6] Usually histopathological examination is key to diagnosis. Management options include expectatnt management for small asymptomatic cases, surgical excision for troublesome or large nodules, hormonal medication that help to shrink the nodules like oral contraceptive pills, aromatase inhibitors like letrozole, or gonadotrophic-releasing hormone (GnRH) analogues like goserelin acetate (GnRH agonist).[5–7]

CASE PRESENTATION

Patient history

Mrs. OO, a 41-year-old Para 0^{+0} referred to the Department of Obstetrics and Gynaecology, Redeemer's Medical Centre, Redemption Camp, Mowe, Ogun State, Nigeria; with recurrent huge abdominal swelling of 4 years duration. Lower abdominal swelling was of insidious onset following an abdominal myomectomy done four and half years previously, for huge uterine fibroid; gradually increasing in size to a huge abdominal mass occupying the whole abdominal cavity. Associated inability to conceive, recurrent lower abdominal pain, menorrhagia and weight loss. No other swellings in any other parts of her body. There was no dysmenorrhoea or intermenstrual bleeding. No history of Papanicolaou smear done. There was no family or personal history of gynaecological cancers. She has a family history of uterine fibroids. She did not take tobacco products or alcoholic beverages.

Clinical examination

General examination revealed short stature (height about 1.4m), asthenia and mild pallor. Abdominal examination revealed a midline anterior abdominal wall suprapubic scar extending above the umbilicus, huge irregular nodular abdominal mass extending from the pelvis to the xyphysternum and costal margins bilaterally, firm, non-tender and minimally mobile. Vaginal examination revealed normal vulva

and vagina, with cervix displaced postero-superiorly but otherwise grossly normal, huge uterus 36 weeks size, and free adnexae bilaterally.

Management

Mrs. OO had preoperative investigations (packed cell volume – 26%, otherwise normal full blood count, electrolyte urea and creatinine assay, urinalysis and negative viral markers) which were otherwise normal. Abdominopelvic ultrasound scan and computerized tomographic scan of the abdomen showed the huge uterine masses with no other pathologies in the abdomen. She was counselled on the need for total abdominal hysterectomy preferably; or repeat abdominal myomectomy and invitro fertilization afterwards, since she was a nulligravida. She opted for the latter. She was transfused with 1 unit of blood peri-operatively and second unit post-operatively. A repeat abdominal myomectomy, incidental appendectomy and excision biopsy of intestinal and mesenteric nodules was done under general anaesthesia with endotracheal intubation.

Bladder was catheterized, routine cleaning and draping was done. Previous scar was excised and wound deepened to peritoneal cavity separating adhesions bluntly and sharply, and noting the findings of multiple large and small uterine fibroids subserous, intramural and submucous (about 34 nodules in all ranging between about 0.5 to 16cm in widest diameter). Also disseminated tiny (0.1 to 0.5cm in widest diameter) well-defined fibrous nodules on the leaves of the broad ligament (also mesosalpinges and mesoovarii)- Fig. 1, on the mesentry of the intestines, mesoappendix and all over the visceral peritoneum of the intestines - Fig. 2, and appendix, also all over the parietal peritoneum. Fibroids were enucleated one after the other, and cavities closed in layers and haemostasis secured. Appendectomy was done, excising a nodule from the mesoappendix and 2 other nodules were enucleated from the antimesenteric surface of the small intestine. All specimen was sent for histology. Anterior abdominal wall was closed by mass closure and skin by matress sutures using nylon 2 – 0. A size 8 intracervical catheter was inserted intrauterine and the balloon inflated with 5ml of water for injection (to be retained for 10 days).

Her postoperative recovery was satisfactory. She had peri-operative intravenous antibiotics and analgesics for 72 hours after surgery. She was discharged home on the fifth post-operative day on oral broad spectrum antibiotics and analgesics. She was given a two-week gynaecological clinic appointment. At the gynaecology clinic visit, she had no complaints. She presented her histopathological report, after which she was commenced on goserelin acetate 3.6mg monthly for 3 months. She was also planned for IVF after completing the 3 months of goserelin acetate.

Histopathological findings

MACROSCOPY: a) Uterine nodules: Received 34 nodular greyish white masses altogether weighing 910g and ranging from 1.0 x 1.0 x 1.0 to 12.0 x 9.0 x 9.0cm. Cut sections show greyish white whorled surfaces. b) Peritoneal nodules: Received 3 greyish white nodules ranging from 0.2 x 0.1 x 0.1cm to 0.5 x

0.5 x 0.5cm. Cut section shows greyish white surfaces. c) Appendix: Received an appendix tissue measuring 4.5 x 0.5cm. There is an attached mesentery having a yellowish white nodular surface and moderate serosal congestion.

MICROSCOPY: Sections of the uterine and peritoneal nodules are similar and show a benign mesenchymal neoplasm composed of proliferating bundles of mature smooth muscle cells disposed in interlacing fascicles and whorls – Fig. 3. Sections of the appendiceal mesenteric nodule show leiomyomatous nodule with focal embedded islands of endometrial-like glands and stroma. Also seen in the appendix section are unremarkable mucosal glands and hyperplastic mucosal follicles with reactive germinal centres and serosal congestion. No focus of malignancy is seen – Fig. 4.

DIAGNOSIS: Uterine, peritoneal and mesoappendiceal nodules – Leiomyomata; with Leiomyomatosis Peritonealis Disseminata and reactive lymphoid hyperplasia of the appendix with endometriosis.

DISCUSSION AND CONCLUSIONS

Leiomyomatosis peritonealis disseminata (LPD) is a rare, typically benign condition characterized by multiple smooth muscle nodules proliferating along peritoneal and abdominopelvic surfaces. Just about 200 cases have been reported worldwide since the condition was first described in 1952.[1–3]

The exact pathogenesis of leiomyomatosis peritonealis disseminata remains unclear, though hormonal stimulation, particularly estrogen and progesterone, and iatrogenic factors are strongly implicated.[2,3] Several proposed mechanisms include stem cell metaplasia, genetic predisposition, and implantation of fragmented leiomyoma tissue during laparoscopic morcellation.[3,4] A strong association exists with prior uterine surgery; in one clinical series of 13 patients, 11 had a surgical history related to uterine leiomyoma.[5] LPD predominantly affects women of reproductive age, though postmenopausal women and men have also been reported. Familial clustering with autosomal-dominant inheritance has been suggested.[2–4] The case of LPD reported in South-East Nigeria had total abdominal hysterectomy and bilateral salpingo-oophorectomy, with meticulous excision of peritoneal nodules. Patient developed type 2 diabetes mellitus (T2DM) about one month after surgery, raising the question of a possible association between LPD and T2DM.[8] Another case reported in Nigeria had a history of prolonged exposure to combined oral contraceptive pills (for six years) which was suspected to be related to the disorder in this case, since LPD is associated with high levels of exogenous or endogenous ovarian steroids.[11]

Preoperative diagnosis is difficult because LPD sometimes mimics peritoneal carcinomatosis, leiomyosarcoma, and gastrointestinal stromal tumors on imaging.[6] Most patients are asymptomatic or present with non-specific abdominal pain and vague discomfort. The index patient presented with recurrent uterine fibroid with menorrhagia. CT, MRI, and PET imaging help delineate disease extent but cannot definitively exclude malignancy.[7] Histopathological examination remains the gold standard, with immunohistochemistry showing positivity for smooth muscle actin, estrogen receptor, and progesterone receptor, and a low Ki-67 proliferation index confirming benignity.[4,6,7]

No standardized treatment guidelines exists due to LPD's rarity, and the fact that diagnosis may first be made during surgery as occurred in the index patient where the disseminated tiny peritoneal nodules which were initially missed during the pre-operative radiological investigations, were first seen at surgery. Surgical resection is the mainstay for symptomatic or complicated cases. GnRH agonist therapy represents a medical alternative, particularly for recurrent or hormonally sensitive disease. This management option was employed for the index patient, after repeat abdominal myomectomy, excision biopsy of nodules and appendectomy. The choice of myomectomy was a limitation, and was taken due to the nulliparity of the patient and her strong desire for invitro fertilization (IVF) afterwards. This was supported by the fact that peritoneal nodules in the index patient were very tiny ranging from 0.1 to 0.5cm in widest diameter, which are more likely to be sensitive to adjuvant hormonal therapy; also the high index of suspicion of LPD during findings at surgery and its largely benign nature. Cytoreductive surgery with hyperthermic intraperitoneal chemotherapy has been explored for aggressive recurrence.[12–14] Although prognosis is generally favorable, recurrence occurs in a subset of patients and rare malignant transformation to leiomyosarcoma or endometrial sarcoma has been documented, necessitating long-term surveillance.[14]

In conclusion, leiomyomatosis peritonealis disseminata is a very rare benign condition, thus, a high index of suspicion is required preoperatively in women with recurrent uterine fibroids presenting with the other typical risk factors for the condition. Such disseminated nodules excised during surgery should be subjected to histopathologic assessment to clinch the diagnosis.

Abbreviations

AP
USS–Abdominopelvic Ultrasound Scan
COCP
Combined oral contraceptive pills
C/S
Caesarean section
CT Scan
Computerized Tomographic Scan
EUCr
Electrolytes urea and creatinine
FBC
Full blood count
GnRHa
Gonadotrophin Releasing Hormone analogues
H & E
Haematoxylin and Eosin stain
IVF

In–vitro Fertilization
Ki67
Kiel 67
LPD
Leiomyomatosis peritonealis disseminate
MRI
Magnetic Resonance Imaging
PCV
Packed Cell Volume
PET
Positron Emission Tomographic Scan
RMC
Redeemed Medical Centre
T2DM
Type 2 Diabetes Mellitus
TAH
Total abdominal hysterectomy
TAUSS
Transabdominal ultrasound scan
TVUSS
Transvaginal ultrasound scan

Declarations

Ethics approval and consent to participate –

• not applicable

Consent for publication -

• Patient gave written informed consent for her personal and clinical details along with the identifying images to be published

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Author Contribution

- N. O. – did the surgery, literature review and write-up of the manuscript- O. N. O. – managed patient pre-operatively, postoperatively and follow-up, obtaining consent for publication- J. I. N. – did all the

histopathological examination, evaluation and reportingAll authors reviewed the manuscript

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Figures



Figure 1

Intraoperative findings of leiomyomatosis on the broad ligament including mesosalpinges and mesoovarii.

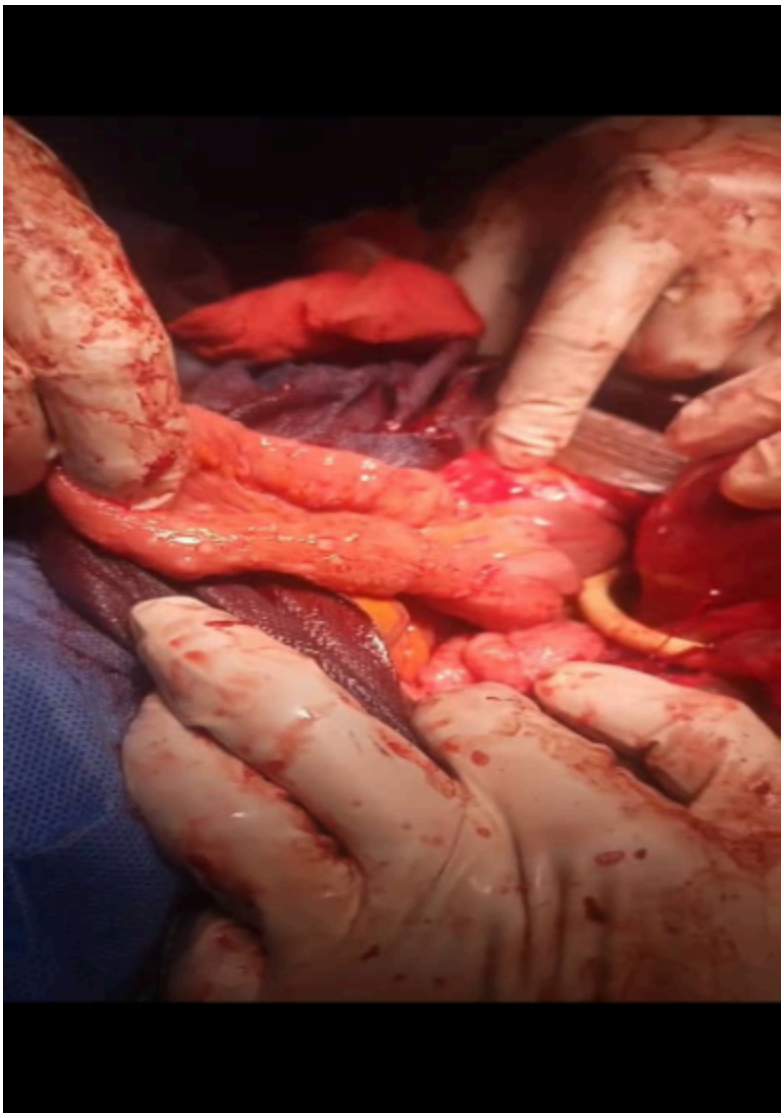


Figure 2

Intraoperative findings of leiomyomatosis on the intestines and mesentry.

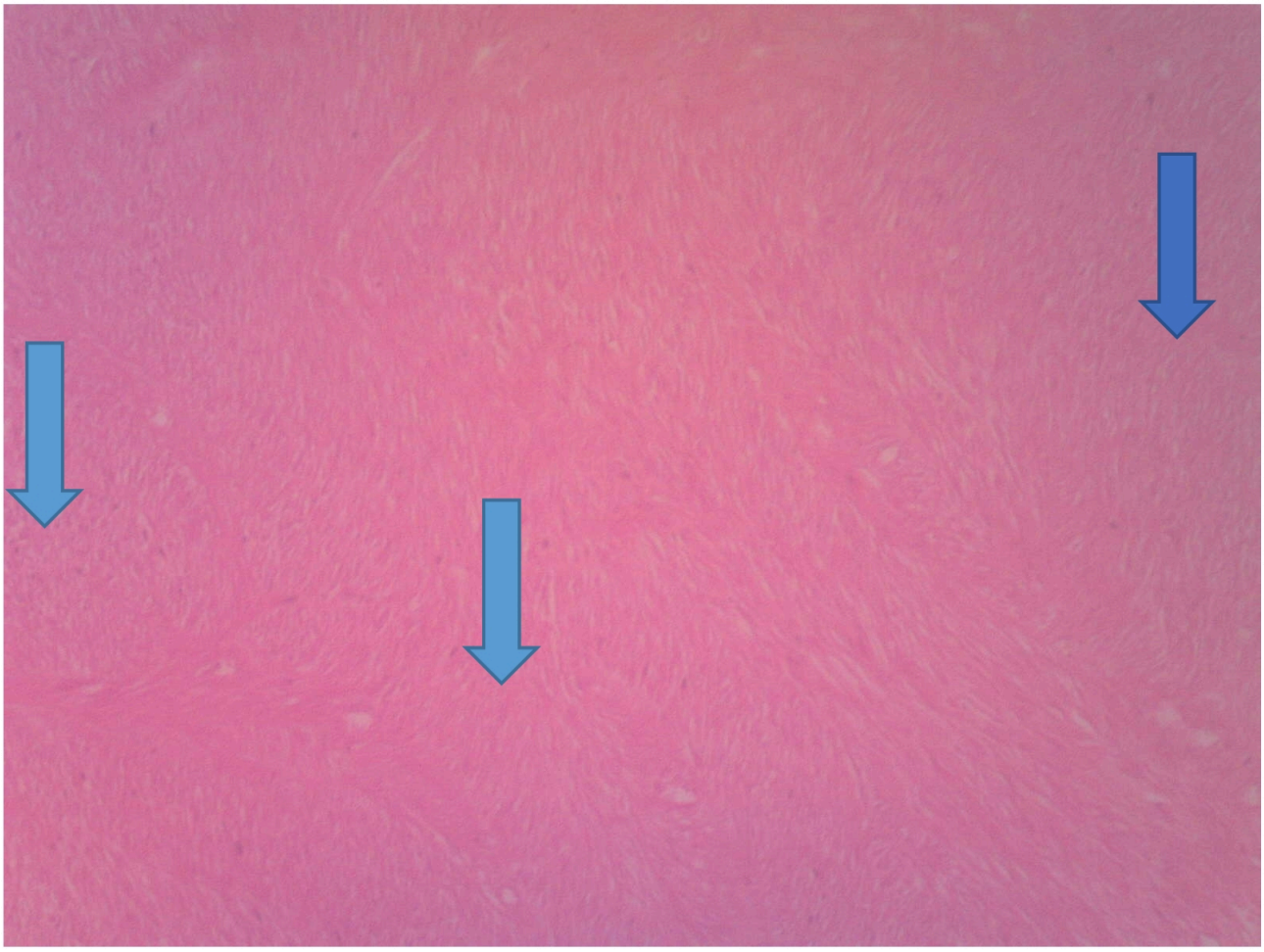


Figure 3

H & E @ x 100 showing uterine nodules composed of proliferating bundles of mature smooth muscle cells disposed in fascicles and whorls (arrows) in keeping with leiomyoma

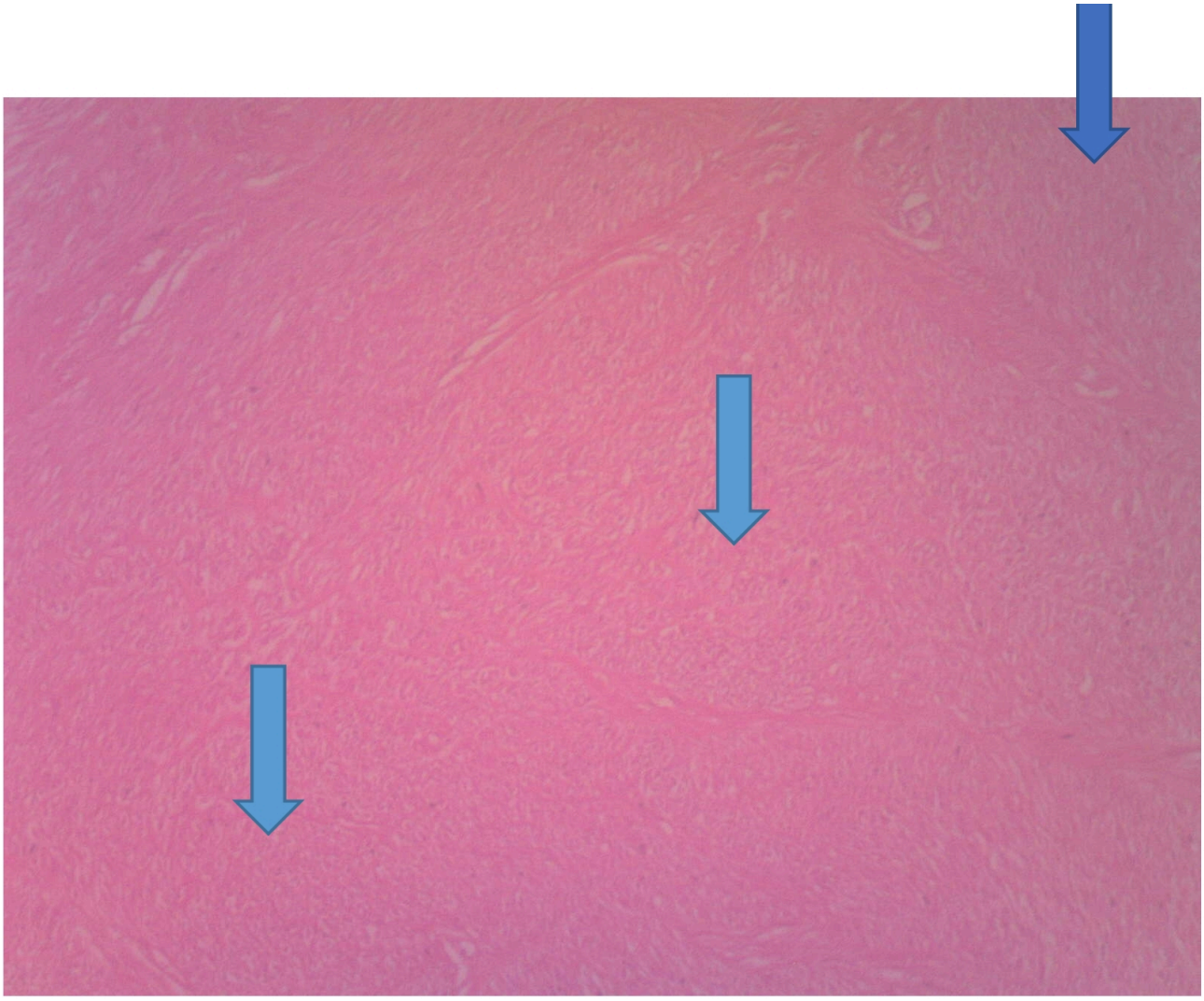


Figure 4

H & E @ x 100 showing Peritoneal and mesoappendiceal nodules composed of proliferating bundles of mature smooth muscle cells disposed in fascicles and whorls (arrows) in keeping with Leiomyomatosis Peritonealis Disseminata

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