CASE REPORT





Port Site Leiomyoma and Leiomyomatosis Peritonealis Disseminata Following Uncontained Uterine Power Morcellation: A Case Report

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Introduction

Leiomyomatosis peritonealis disseminata (LPD)/parasitic leiomyoma (PL) is a rare variant of smooth muscle nodules occurring outside the uterus. According to FIGO classification, parasitic leiomyoma is of type 8, Kelly and Cuccess in 1909 first described parasitic leiomyoma. Extra uterine leiomyomas are rare, present with unusual growth pattern or occur in an unusual location that make their identification more challenging both clinically and radiologically. The clinical presentation is nonspecific and depends on the site of recurrence. Most of these patients are asymptomatic and if symptomatic abdominal swelling/pelvic mass, pressure or abdominal distension, are the manifestations. A fibroid away from the uterus with a history of laparoscopic myomectomy/hysterectomy with power morcellation gives a clue of LPD/parasitic leiomyoma.

Surgical excision is the main treatment.

Case Report

A 28-year-old nulligravida consulted us with a complaint of mass in the upper abdomen, noticed 5 months ago. She underwent laparoscopic myomectomy and power morcellation for a very large fibroid (unknown dimensions) seven

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years ago in 2015. Examination revealed a swelling beneath the supra umbilical trocar site. Subsequent ultrasound and magnetic resonance imaging (MRI) done during evaluation by us during February 2022 demonstrated multiple tumors of varied sizes all over the abdomen that included the port site (Fig. 1).

Multiple lobulated well-defined space occupying lesions that varied in size with MR signal pattern of myomata (T2 hypointense, T1 Isointense, DWI low signal) were noted in the abdominal cavity in different locations (intraperitoneal, anterior abdominal wall and portside entry) attached to the mesentery, peritoneum and anterior abdominal wall with characteristics features of parasitic leiomyomata, mapped in the pelvic cavity, 43 mm largest diameter on the right side, 31 mm, 21 mm and, 66 mm on the right peritoneal, anterior abdominal wall with cystic degeneration spread over the transverse abdominal muscle (9.4 cm). Portside entry at umbilicus 49 × 66 mm, 55 mm at the mesenteric attachment in supramesocolic compartment, 49 mm in left paracolic gutter, 21 mm along right round ligament was noted. Vascularity from the peritoneum (Table 1).

Majority of the myomata were predominantly T2 dark/diffussion weighted imaging (DWI) dark/apparent diffusion coefficient (ADC) low (between 0.9–1.1) consistent with benign morphology: T2 dark DWI one myoma in right anterior abdominal wall showed central T2 hyperintensity DWI bright with ADC value of 1.6–1.9 suggesting T2 shine through and cystic degeneration.

Therapeutic intervention: an operative laparoscopy was performed under general anesthesia GA. Multiple white gray nodules of varying sizes at different locations were noted. Twenty-one nodules were excised. All the tumors removed intact through a small minilap suprapubic incision, thus avoiding morcellation. Histopathological examination of the tissue was consistent with fibroids (Fig. 2).

Of the 21 fibroids, the largest was 11.9 cm, anterior abdominal wall fibroid showed cystic degeneration splaying the transverse abdominal muscle. Most of the fibroids



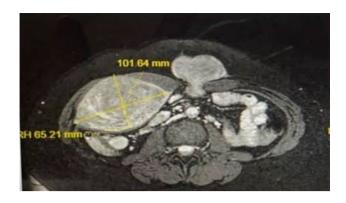


Fig. 1 Magnetic resonance imaging features of the tumour

Table 1 Location of the fibroids in our case report

Location	Number
Right adnexa 43 mm	1
Transverse abdominus muscle 11.9 cms	1
Left paracolic gutter 49 mm	1
Port site 66×49 mm	2
Above the round ligament on the right side 21 mm	2
Anterior to the sacral promontory	2
Supra mesocolic mysentry 55 mm	2
Right lateral peritoneal wall 31,21,66 mm	3
In other areas	7

derived the vascularity either from the peritoneum/mesentry/omentum.

There were no uterine leiomyomas and the previous myomectomy scar was intact. Blood loss was very minimum intraoperatively. Post-operative recovery was uneventful.

Multiple fibroids varied in size from 0.6×0.4 to 11.6×11.9 cm, serial sections across all the fibroids were solid grey white and grey brown and whorled with focal myxoid areas macroscopically.

Microscopic examination revealed leiomyomas with fascicles of smooth muscle cells with oval nuclei and fine chromatin (Fig. 2). Intervening hypocellular foci was noted. There was no evidence of malignancy.

Discussion

LPD is a rare late sequel of uterine leiomyomas, first described in 1952 by Wilson and Paele. It can affect women in their reproductive age, premenopausal and sometimes in postmenopausal women. The etiology and pathophysiology of LPD can be either primary or iatrogenic, the latter being more common due to uncontained power morcellation of fibroids, which augments the potentiality of tumor implantation and dissemination.

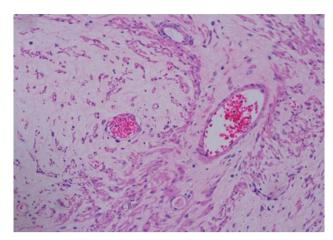


Fig. 2 Histopathological features of the tissue confirming fibroids (Hematoxylene and eosin 10x)

LPD originates from metaplasia of submesothelial, multipotential mesenchymal cells and associated with exposure to high endogenous or exogenous estrogens, as in prolonged usage of contraceptive pills and pregnancy. In LPD, tumor cells have both estrogen and progesterone receptors. In addition, autosomal dominant model with varying level of penetrance has been reported in the literature, explaining the occurrance of familial clustering.

Due to uncontained laparosocopic myomectomy/hysterectomy procedures the LPD/Parasitic leiomyoma cases are increasing. Kimberly and his colleagues reported 12 cases of parasitic fibroids with a history of previous morcellation procedure. Lu et al. presented 6 cases of such tumors. All of them had history of myomectomy/hysterectomy with power morcellation.

A retrospective study over 3 years by Gaspare and associates showed the development of PL after power morcellation. Of 425 cases, 0.9% developed PL. It was concluded that uncontained morcellation is a risk factor for developing LPD/PL. Thus came In-bag morcellation of the fibroids to minimise spillage. Table 2 describes the case reports of LPD reported by various authors excluding the 37 cases listed by Huang et al. A careful inspection and thorough washing of the abdomen and pelvic cavities should be done after the procedure.

Majority (93%) of PL occur in pelvis. Many authors reported LPD/PL but rarity in our case is having a leiomyomas in the abdomen and pelvic areas along with the port site and on the large intestine. Dashraath et al. reported the mass on greater omentum. Concomitant occurrence of uterine leiomyomas and LPD have been reported. In our case, uterus, both fallopian tubes, and ovaries were absolutely normal with an intact previous myomectomy scar.

Intracapsular myomectomy, the removal of fibroids from its pseudocapsule, may prove a promising surgical



Table 2 Reported cases of port-site implantation of parasitic leiomyoma

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Author	Age of the patient (years)	Parity	History of morcellation	Previous myomectomy & morcellation/gyn surgery	Adjuvant therapy	Time after previous sur- gery (months)	Treatment	Pathology	Outcome
Ostrzenski et al. 1997	43	I	None	I	Combined oral contraceptive	2	Local removal	Leiomyoma	N/A
Moon et al. 2008	31	G2P2	Yes	Laparoscopic myomectomy after moreellation—3 years	None	36	Recurrence was treated with local surgical excision	Leiomyoma on both occa- N/A sions	N/A
Ono et al. (abdominal wall and uter- ine myoma) 2010 Japan	37	G2P2	Yes	1	None	1	Local removal	Extensive calcification,degenerative Leiomyoma	N/A
Huang et al. 2014. Taiwan	27	Nulliparous	1	Yes; laparoscopic myomectomy with morcellation for a submucosal myoma	None	2 8	Laparoscopic excision	Leiomyoma	At the time of reporting, patient was under follow-up
Yorita et al. 2016. Japan	47	nulliparous	Yes	None	None		Extirpation of the pedunculated uterine mass and cystectomy of the left ovarian lesion	Torsion of the pedicle of the ovarian tumor, leiomyoma with red degeneration.	Uneventful
Salih et al. 2017. Iraq	46	N/A	N _O	No	None	I	Laparotomy	Leiomyoma	No complaints after 3 months follow-up
Clarke et al. 2017. USA	36	G2P2	Yes	Yes. Lapa- roscopic myomectomy six years ago. Patient had two successful pregnancies	None	72	Laparoscopic myomectomy	Leiomyoma	No immediate complications, long term follow-up details not available
Kirplani et al. 2018. India	48	P1, live birth 1	Yes	Hysterectomy -for fibroid -3 years ago	None	36	Local resection	Leiomyoma	Recurrence after 2 years, removed and stable



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Table 2 (continued)

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Author	Age of the patient (years)	Parity	History of morcellation	Previous myomectomy & morcellation/gyn surgery	Adjuvant therapy	Time after previous sur- gery (months)	Treatment	Pathology	Outcome
Oindi et al. [1]. Africa	47	P3+0	Yes	Laparoscopic myomectomy 6 years ago	None	36	Total abdominal hysterectomy, excision of the mass	Leiomyoma	Uneventful, no recurrence after 6 months of surgery
Tan et al. 2019. Singapore	37	N/A	Yes	Yes, laparoscopic myomectomy with unconfined power morcellation—3 years ago	Leuprorelin depot injec- tions—to minimize DPL	36	Local removal	Leiomyoma	After 2 years post surgery, intra-abdominal and pelvic recurrences on CT. Patient is on observation as patient not willing for resurgery
Lee et al. (Recurrent parasitic fibroids). 2019. Singapore	2	PI	Yes	Uncomplicated laparoscopic myomectomy 8 years ago	None	96	Laparoscopic myomectomy initial. Local exci- sion for recurrence (umbilical mass)	Consistent with leiomyoma on both occassions	Recurrence within a month. Post excision of re-recurrence (umbilical mass) -uneventful at 1 year follow-up
Tasdemir et al. 2019. Turkeye	33	GIPI	Yes	Yes, 3 years ago, Laparoscopic myomectomy	None	36	Laparoscopic myomectomy and a power morcellation. Local excision for recurrence		Re-recurrence at one year
Javali et al. 2019. India	30	PIL1	Yes	Laparoscopic myomec- tomy—3 years ago-for cervical fibroid. LSCS—one year ago	None	36	Excision of the mass	Leiomyoma	Uneventful at month 6 visit
	41	Nulligravida	Yes	Laparoscopic myomec- tomy—6 years ago-for fundal fibroid	None	72	Total laparoscopic hysterectomy with open myomectomy	N/A	N/A



Table 2 (continued)

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Author	Age of the patient (years)	Parity	History of morcellation	Previous myomectomy & morcellation/gyn surgery	Adjuvant therapy	Time after previous sur- gery (months)	Treatment	Pathology	Outcome
Kai et al. [2]. Japan	36	Primigravid	Yes	N/A	Combined oral contraceptive six cycles of gonadotropin-releasing hormone agonists (GnRHas)—before consulting the authors	45	Laparoscopic myomectomy and left ovarian cystectomy. Fibroids removed with power morcellation for the initial one. Abdominal myomectyomy for the recurrence	Leiomyoma with low cell density & prominent hyalinization	Recurrence after 3 yrs 9 months, which was operated, no recurrence was noticed at 17 months postsurgery
Adyagi et al. 2020. Japan	30	Primigravida	Yes	Laparoscopic myomectomy with power morcellation and ovarian cystectomy – 7 years ago	Oral contraceptives	48	Abdominal myomectomy	Leiomyoma	N/A
Pai et al. 2020. Taiwan	42	G1P1		Laparoscopic myomectomy, many years ago	None	N/A	Laparocopy and excision	Leiomyoma	N/A
Ozlurk et al. 2021. Turkeye	45	G4P0C4 (Nullipa-rous)	Yes	Yes, Myomectomy eight years ago	None	96	Local removal	Numerous myoma, Leio- myoma	N/A
Smyka et al. [3]. Poland	. 31	₹ Z	Yes	N. N	None		Diagnosed during investigation for patency of fallopian tubes, for infertility work-up Laparoscopic myomectomy	Leiomyoma	N/A
Roh et al. [4]. South Korea	50		Yes	Yes. Laparoscopic myomectomy six years ago		72	Surgical excision	Leiomyoma	No complications
Present case 2022. India	78	Nulligravida	Yes	Yes, Laparoscopic myomemcectomy in	None	72	Local excision of all fibroids	Leiomyoma	Uneventful and- under follow-up

N/A = Not available, DPL = Disseminated peritoneal leiomyomatosis



technique, not only for uterine myometrial healing but also in preventing LPD. A pseudo-capsule is defined as an anatomic structure surrounding the fibroid and separating it from the normal myometrium, which has a high angiogenic potential derived from growth factor iatrogenic damage and spread of pseudo capsule by power morcellation could promote implantation of minced leiomyoma fragments at an ectopic site.

Management is generally resection and excision, which can be by open, laparoscopic or robotic procedure. In this case, to avoid morcellation and further recurrence, a small suprapubic incision was given, and all the fibroids were removed intact.

Conclusion

Port site leiomyoma and LPD are a rare specific, mostly non-malignant sequel following myomectomy and uncontained power morcellation. This can be reduced but not eliminated by the in-bag morcellation.

Clinicians should councel the patient regarding the risk and benefits of laparoscopy with/without in-bag morcellation. The strategy in advanced laparoscopic surgeries is meticulously following the principles of surgery and contained morcellation, which minimizes the probability of LPD.

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Declarations

Conflict of Interest Nil.

Ethical Approval Not required for reporting individual cases.

Human or Animal Consent Nil.

Informed Consent Written informed consent obtained from the patient.

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