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Case Report

Giant uterine leiomyoma: a case report with literature review

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ABSTRACT

Uterine leiomyomas are one of the most common entities encountered in routine gynaecological practice; however, the giant uterine myomas are very rare and can often create a diagnostic dilemma and therapeutic challenge owing to their size, non-specific clinical presentation and degenerations. Here, in this article we review the literature on giant uterine leiomyomas and report one such case in a 38-year-old woman who presented with the complaints of vague abdominal lump, enlargement of abdomen, dysmenorrhea, lower abdominal and pelvic pain since last 2 years and a 6-month history of increased frequency of micturation. Physical examination and radiological investigations suggested a giant abdominopelvic mass, probably a uterine or an ovarian malignancy. An exploratory laparotomy was performed followed by total hysterectomy. Grossly, the specimen was a 15.2 kg uterine leiomyoma measuring 18x18x13 cm in size. Histologically, the benign leiomyoma showed cystic degeneration. The patient's post-operative and follow-up period was uneventful.

Keywords: Cystic degeneration, Giant tumor, Leiomyoma, Uterus

INTRODUCTION

Uterine leiomyomas are the commonest benign tumors of the female genital tract, arising from the smooth muscle cells of the myometrium.¹ The chances of their occurrence increases during the reproductive period and usually decreases after menopause.² They are found in more than half of the women over the age of 35 years but can also be present in adolescents.³ Their exact pathogenesis is still debatable, however hormonal stimulation by estrogen, progesterone and other growth factors plays a pivotal role. Many risk factors have also been implicated for their occurrence like family history, nulliparity, black race, obesity, meat consumption and hypertension. Based on their location they can be categorized as intramural, submucosal and subserosal. Leiomyomas may appear as single or multiple and can be of varying sizes.⁴ Most of the patients have small uterine leiomyoma. However, giant ones do exist and are extremely rare.⁵ They are usually asymptomatic, but large tumors often produce abnormal bleeding, pelvic

discomfort (pressure or pain), dysmenorrhea, infertility, frequent urination, constipation, "myomatous erythrocytosis syndrome", Pseudo-Meigs syndrome or preterm labour.⁶ We herein describe an interesting case of giant uterine leiomyoma with degenerative changes along with review of the pertinent world literature so as to create awareness among the dealing clinicians about this enigmatic entity.

CASE REPORT

A 38-year-old illiterate woman from a rural background presented to gynaecological outpatient department with the complaints of vague abdominal lump, enlargement of abdomen, dysmenorrhea, lower abdominal and pelvic pain since last 2 years and increased frequency of micturation from last 6 months. There was history of recent weight gain and loss of appetite. She had 2 children and her last child birth was 5 years back. Her medical history was non-contributory; she had no major illness or any previous surgical procedures. Her family

history was not significant. On general examination, her vitals were normal except a protuberant abdomen was seen. On abdominal examination, her abdomen was circumferentially distended by a huge abdominopelvic mass, which was non-tender, firm in consistency and dull on percussion. Per speculum examination revealed normal external genitalia and uterine cervix. On bimanual examination, fornices were full and a very large, firm, mobile, central mass that filled the entire pelvis and abdomen was felt. It was difficult to specify the origin of the mass.

Abdominal ultrasonography confirmed the presence of a huge heterogeneous soft tissue mass arising from the pelvis and occupying almost the entire abdomen, the uterus was poorly visualized. Contrast-enhanced computed tomography (CECT) of the abdomen and pelvis revealed a large solid-cystic mass in the pelvis extending into the abdominal cavity. The mass appeared to be arising from the uterus. The right ovary and fallopian tube were normal, however the left sided adnexa was not visible. The lymphnodes were not enlarged. Her routine hematological, microbiological and other biochemical tests including serum CA 125 (cancer antigen 125) levels and pap smear were within normal limits.

Considering the size of the mass and suspicion of a uterine or an ovarian malignancy, the patient underwent laparotomy. On exploration, bilateral ovaries and fallopian tubes were normal and hence, a total abdominal hysterectomy was performed. The specimen was sent for histopathological examination. On gross examination, a total hysterectomy specimen was received measuring 25x19x15 cm in size. Cervical length measured 3 cm and cervical diameter measured 3.5 cm. External surface of the uterus showed few congested blood vessels. On probing, endocervical canal was patent. It weighed 15.2 kg (Figure 1).

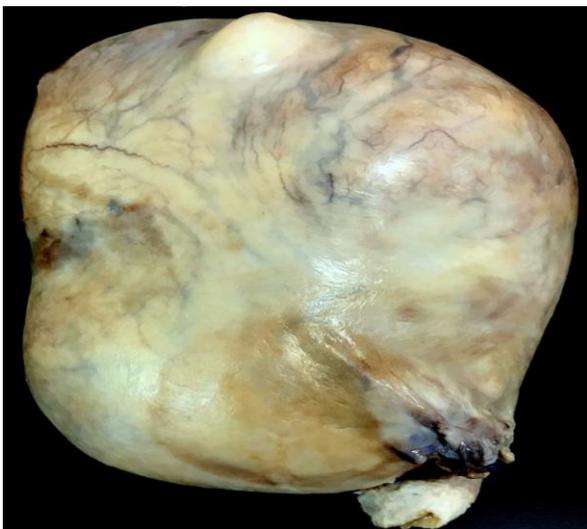


Figure 1: Postoperative specimen of a huge uterus with cervix.

Cut surface showed a large, grayish-white, sharply circumscribed, firm intramural fibroid measuring 18x18x13 cm in size with characteristic “raw silk” and whorled pattern. Few cystic areas were also evident. No haemorrhage or necrosis was seen. Endometrial canal was compressed, and its thickness measured 0.2-0.3cm (Figure 2).



Figure 2: Cut section of the hysterectomy specimen showing multiple gray-white masses with whorled appearance and small cystic areas in between.

Microscopic examination revealed features of a benign leiomyoma with cystic degeneration (Figure 3). The cervix was unremarkable. After the surgical procedure the patient was hospitalized for 10 days and the postoperative period was uneventful. Her symptoms resolved and there were no fresh complaints on 2 month follow-up.

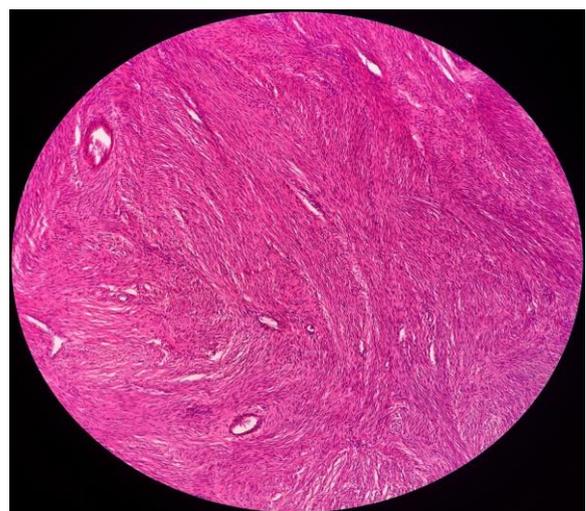


Figure 3: Tumor composed of mixture of spindle shaped smooth muscle fibres and fibrous tissue in varying proportion cut transversely and longitudinally (H and E, x10).

DISCUSSION

Giant uterine leiomyomas are uncommon benign neoplasms and are those which weigh more than 11.4 kg or have a diameter which is more than 17 cm or

dimension 33x28x22 cm.^{2,7} In literature, till date less than 100 cases of giant uterine myomas have been documented worldwide. The largest uterine fibroid ever reported weighed 63.3 kg, which was removed postmortem in 1888.

Table 1: Giant uterine leiomyoma cases reported in the literature from the year 1800 to 2010.

First Author (Reference)	Clinical presentation	Weight/size of leiomyoma	Other pathological features and associated conditions	Outcome
Hunter SH ⁸	AS and pressure symptoms	63.6 kg	NA	Post-mortem
Behrend ⁸	AS	60.7 kg 60x58x29 cm	Simulating ovarian cyst having both solid and cystic areas	Died of pneumonia after 48 hours of operation
Singhabandhu B ⁸	NA	45.4 kg	NA	Survived
Jonas HS ⁹	NA	29.48 kg	NA	NA
Evans AT ¹⁰	NA	11.4 kg	NA	NA
Mazzocconi G ¹¹	NA	NA	Pedunculated fundus uteri	Uncomplicated
Ozsaran AA ¹²	AM and plethora	14.2 kg	Subserous myoma associated with erythrocytosis	Uneventful
Djelmis J ¹³	NA	25 kg	NA	NA
Oelsner G ¹⁴	Severe pulmonary hypertension and respiratory failure	40 kg and 43.2 kg	NA	Uneventful
Nguyen-Duc H ¹⁵	AM, menorrhagia and severe anemia	NA	Giant leiomyoma in a 15-year-old teenager	Uneventful
Costa BL ¹⁶	AM, constipation and respiratory difficulty	12.4 kg	NA	Uneventful
Perez M ¹⁷	AD and WL	27 kg 65x54x25 cm	Uterine myoma with myxomatous degeneration, calcification and chondroid metaplasia	Uneventful
Panayotidis C ¹⁸	Pelvic mass and pressure symptoms	4.5 kg 31x26x15 cm	Fetal shaped fibroid uterus (multiple fibroids)	Uneventful
Nappi L ¹⁹	AD	27.7 kg	Bilobed giant myoma	Uneventful
Karim T ²⁰	AM and AD	3.2 kg 25x15x10 cm	Giant fibroid in a 16-year-old adolescent girl	Uneventful

NA: Not Available, AD: Abdominal Distension, AS: Abdominal swelling, AM: Abdominal mass, AE: Abdominal enlargement, WL: Weight loss

On the other hand, the largest one ever removed from a patient who survived the procedure weighed 45.4 kg. This fibroid was reportedly the 34th weighing more than 18.2 kg since 1878.⁸

Table 1 summarizes the clinicopathological features of giant uterine leiomyomas which were reported from the year 1800 till 2010. On further extensive reviewing the cases since the year 2011 till date (Table 2 and 3) it was

noted that there was a surge in the number of patients being reported with giant uterine leiomyomas.

The uterine fibroids have an unremarkable potential to grow to an incredible extreme size before disabling the patient or producing appreciable symptoms. The only limit on size seems to be the capability of the host to bear it. This is owing to the relatively large volume of the abdominal cavity, the distensibility of the anterior abdominal wall, and the slow growth rate of this tumor.

Table 2: Literature review of giant uterine leiomyoma cases reported from the year 2011 to 2014.

First Author (Reference)	Clinical presentation	Weight/size of leiomyoma	Other pathological features and associated conditions	Outcome
Steward RG ²¹	AE with history of uterine fibroids	11.618 kg 31x26x14 cm (largest)	Conglomerate, pedunculated leiomyomata	Uneventful
Savulescu F ⁶	AE, WG, pressure symptoms, dysmenorrhea, dyspareunia, menorrhagia, lower abdominal and pelvic pain, frequent urination and constipation	18.1 kg 33x28x22 cm	Bilobed solid tumor with multiple subserosal fibroids	Uneventful
Murtaza B ²²	AM, dull pain in lumbar region, constipation, WL	7.4 kg 55x40x35 cm	Pedunculated subserosal fibroid with bilateral hydronephrosis	Uneventful with complete regression of bilateral hydronephrosis after 2 months of surgery.
Öndeş B ²³	AD	25 kg 33x28x23 cm	Few cystic and ulcerated areas	Postoperative wound dehiscence
Al-Obaidi SM ²⁴	AM, AD, constipation, difficulty in initiation of micturition	19.8 kg 60x45x25 cm	Atrophied uterus with fibroid and cystic degeneration of benign ovarian tissue	Uneventful
Alam IP ²	AM, AD, occasional constipation, increased frequency of micturition	9.5 kg	Multiple fibroids, few showing necrosis and few others showing cystic and degenerative changes	Uneventful
Aydin C ²⁵	Lower abdominal pain, AD, recent WG of 25 kg	33x20x18 cm	Multiloculated, cystic, pedunculated leiomyoma with extensive cystic degeneration	Uneventful
Mate S ²⁶	Acute dyspnea and severe acute anemia (due to intratumoral bleeding)	13.5 kg	Pedunculated leiomyoma	Uneventful
Gajewska M ²⁷	AE	25 cm in diameter	Pedunculated leiomyoma with cystic degeneration	Uneventful
Funaki K ²⁸	AD and WG	8 kg 40x40x30 cm	Subserous leiomyoma with cystic degeneration	Uneventful
Konard Wronski ¹	Abdominal pain, AD, constipation	24.2x17.3 cm	Uterine leiomyomata with cystic degeneration	Uncomplicated
Ezugwu EC ⁴	AS, infertility, WL	16.8 kg (total fibroid weight) Largest: 12.4 kg, 32x24 cm	Large conglomerate fibroid masses attached to fundus along with multiple intramural fibroid seedlings.	Uneventful recovery and successful pregnancy seven months following myomectomy.
Moris D ⁷	Progressive constipation, increasing abdominal size, back pain, vague abdominal pressure sensations and urinary frequency	28.1 kg 62x39x21 cm	Solid and nodular	Uneventful

NA: Not Available, AD: Abdominal Distension, AS: Abdominal swelling, AM: Abdominal mass, AE: Abdominal enlargement, WL: Weight loss, WG: Weight Gain

As a result, these patients can present in a physical condition which is unlike that of a typical woman with fibroids, but instead more like that of a much older patient with multiple comorbidities.²¹ However, the giant

neoplasms frequently causes abnormal periods, pelvic pain and pressure effects on lungs, urinary bladder, ureters and other adjacent organs leading to further complications like lower-limb thrombosis as well as renal and respiratory failure.²

Table 3: Giant uterine leiomyoma cases reported from the year 2015 to 2018 (till date).

First Author (Reference)	Clinical presentation	Weight/size of leiomyoma	Other pathological features and associated conditions	Outcome
Kalayci TO ²⁹	Abdominal pain and AD	30x25x17 cm	Pedunculated subserosal leiomyoma with degenerative changes	Uneventful
Yavuz A ³⁰	Menometrorrhagia and AS	9.8 kg	Associated umbilical hernia	Uneventful
Mulayim B ³¹	No symptoms or complaints	3.98 kg 17 cm	Intramural, cellular leiomyoma without secondary changes	Uneventful
Sharma RP ³²	Gradual WG, increasing abdominal size, vague abdominal pressure sensations, dysmenorrhoea, dyspareunia, menorrhagia, lower abdomen and pelvic pain, frequent urination and constipation	20 kg 36.4x28.7x23.3 cm	Fibroid arising from fundus region	Uneventful
Reshmy JR ³³	AD and lower abdominal pain	28x20x10 cm	Leiomyoma with cystic degeneration	NA
Rahman H ⁵	AS, vague abdominal discomfort, lower abdomen and pelvic pain and frequency of urination	11.6 kg 43x32x23 cm	Associated erythrocytosis	Complete recovery after hysterectomy
Carbunaru A ³⁴	AM with left lower limb swelling	4.5 kg 25x21x20 cm	Myoma with hyaline degeneration and associated deep vein thrombosis of left lower limb	Uneventful
Gennaro Della Rossa MN ³⁵	AM	23x19x12 cm	Intramural fibroid with minimal nuclear atypia and low mitotic activity with interstitial edematous degeneration without signs of necrosis	Uterine reconstruction following myomectomy with uneventful recovery
Ramteke S ³⁶	AD, WL, decreased appetite, bleeding per vaginum	6.5 kg 26x23x18 cm	Intramural fibroid	NA
Muller R ³⁷	AE and WL	15.6 kg	NA	Uneventful
Hrgovic Z ³⁸	AD	25 cm diameter	Myoma spontaneously regressed in size 3 years after delivery	Pregnancy outcome favourable
Bartos V ³⁹	AE	8.1 kg 30x30x20 cm	Subserous leiomyoma with regressive and degenerative changes	Uneventful

NA: Not available, AD: Abdominal distension, AS: Abdominal swelling, AM: Abdominal mass, AE: Abdominal enlargement, WL: Weight loss, WG: Weight gain.

Furthermore, on enlargement, uterine fibroids undergo different types of degeneration such as hyaline, cystic, myxoid or red degeneration and dystrophic calcification. Hyaline degeneration is most common (60% cases of leiomyomas) while cystic degeneration occurs in 4% of

such cases.¹ These degenerations arise due to the inadequate blood supply and seem to depend on the degree and rapidity of the onset of vascular insufficiency. However, researchers have found no relationship between any symptoms and the incidence of degenerative

changes.² The diagnostic approach whenever a uterine leiomyoma is suspected is the pelvic examination which is the first step of evaluation in such patients, however, usually the smaller ones are not palpated and it is only the very large myomas which can be felt during examination.⁴ Therefore, imaging studies should be considered as they are helpful in determining the location, number, size and extent of the leiomyomas.²¹

Ultrasonography is preferred as the initial screening tool because it is least invasive and most cost-effective investigation.^{2,6} Computed Tomography (CT) and Magnetic Resonant Imaging (MRI) are quite helpful especially for ruling out any malignant transformation and in differential diagnosis but they have certain limitations. On CT scans, sometimes the leiomyomas are indistinguishable from healthy myometrium unless they are calcified or necrotic while MRI has limited availability and high cost restricting its use.⁶ Tumor markers have also been implicated to play an important role in its diagnosis as based on them the likelihood of malignancy can be ruled out. However, there is no single best modality to diagnose it pre-operatively and usually the giant uterine leiomyomas are diagnosed after laparotomy on histopathological examination. The differential diagnosis includes adenomyosis, hematometra, uterine cancer (carcinoma, sarcoma and carcinosarcoma), ovarian and retroperitoneal cysts or malignancy.^{21,25,27} In the present case study too, it was confused with a uterine or an ovarian malignancy both clinically and radiologically. However, it turned out to be a case of giant uterine leiomyoma with cystic degeneration on histopathology.

The treatment in cases of giant uterine leiomyomas is usually individualized as both the symptoms severity and patients desire to preserve the fertility are very important factors in determining any therapeutic intervention.⁶ The options include expectant management, medical management, surgery, interventional procedures like uterine artery embolization and ablative techniques. Expectant management with observation is done in cases of women with asymptomatic small and large fibroids, since rarely uterine leiomyomas may undergo sarcomatous transformation. Patients with giant myomas present as unusual challenge even for the most experienced gynecologists mainly due to massive blood loss caused by increased vascularity and postoperative possible complications like injury to bowel and urinary tract, infections, haematomas.³ Therefore, the medical management using Gonadotrophin Releasing Hormone (GnRH) analogues has been used to suppress estrogen production, thereby reducing the size of existing myomas and blood loss before the surgery. Uterine artery embolization also aims to decrease intraoperative blood loss, as it causes myoma infarction with very less adverse effects. Surgical treatment includes hysterectomy, myomectomy and myolysis. The removal of giant uterine leiomyomas by total abdominal hysterectomy with or without salpingo-oophorectomy is the traditional

treatment of choice as these leiomyomas may cause infertility and in the presence of pregnancy they may affect the outcome. However, other approaches like vaginal and laparoscopic hysterectomies are associated with less postoperative complications and quicker recovery of the patient as compared to laparotomy /abdominal approach. Myomectomy is performed through laparotomy, laparoscopy or hysteroscopy and is preferred in symptomatic women who refuse hysterectomy or have a desire to retain their fertility. However, it causes significant perioperative morbidity. Myolysis including mono or bipolar cautery, Nd-YAG laser vaporisation or cryotherapy is currently experimental and is associated with risk of recurrence.²

CONCLUSION

Giant uterine leiomyomas are exceedingly rare and can present with variable clinical manifestations. Therefore, this entity should always be kept in mind while dealing with patients with giant abdominopelvic tumors. Ultrasound stays as the initial screening tool for its diagnosis, but most of them are usually diagnosed on laparotomy followed by histopathological examination. Nevertheless, the treatment approach for every patient should be individualized and appropriate multidisciplinary approach with meticulous patient pre and postoperative care is essential for a better outcome in such patients.

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