Case Report

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Rare Tumors
Volume 16: 1–8

DOI: 10.1177/20363613241285089
journals.sagepub.com/home/rtu
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Large subserous uterine leiomyoma presenting as intraabdominal tumor: A case report

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Abstract

Uterine leiomyomas are common benign gynecological tumors due to the overgrowth of uterine smooth muscle. Pedunculated uterine leiomyoma occurs when the mass is in continuity with the uterus with a stalk and may grow either within the uterine cavity or outside of the uterus and may mimic ovarian neoplasms or intraabdominal tumors. Presented is a 28-year-old woman with a progressive abdominal swelling in the past 9 months seen at the surgical outpatient of our facility. Preoperative CT suggested a diagnosis of an intrabdominal cystic. She had laparotomy and was offered myomectomies on account of a large subserous uterine mass arising from the right side of the uterine fundus, small subserous fundal mass, intramural mass in the left side of the fundus and a cervical mass. Histology confirmed multiple uterine leiomyomas with extensive cystic degenerative changes of the large subserous uterine myoma and adenomyosis of the left fundal mass. Detecting the continuity of an abdominal mass even with extensive degenerative changes mimicking a cyst in continuity with the uterus by a pedicle sign on imaging in the absence of ascites should arouse the diagnosis of pedunculated subserosal leiomyoma. This should be further heightened when it is found in association with cervical myoma. Subserous uterine leiomyoma should be considered in a patient of childbearing age with a grossly distended abdomen without obvious evidence of pregnancy or malignancy. Large subserous uterine leiomyoma in an intraabdominal location may present with diagnostic and surgical challenges that require interdisciplinary cooperation.

Keywords

Large subserous leiomyoma, multiple leiomyomas, adenomyosis, uterus, abdominal swelling

Introduction

Leiomyomas or fibroids are the most common benign gynecological tumours that arise from the overgrowth of smooth muscle and connective tissue of the uterus.[1,2] Giant uterine leiomyomas are rare. It is described as giant when it weighs 11.4 kg (25 pounds) or above.[3,4] The largest removed leiomyoma weighed 63.3 kg at post mortem in 1888 and 45.4 kg from a person who survived.[5] It is estimated that less than 100 cases of giant uterine leiomyomas had been documented in literature.[5] The prevalence of uterine leiomyoma is increased during reproductive phase of life and rapidly decrease following menopause and highlighted the importance of hormonal factors as the cause

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for the neoplasm.[5,6] However, the underlying pathogenesis of their development remains unclear with several risk factors such as positive family history, genetic alterations and lifestyle factors identified.[2]

Pedunculated uterine leiomyoma occurs when the mass is in continuity with the uterus with a stalk and may grow either within the uterine cavity (submucosal) or outside of the uterus (subserosal) and the later may simulate ovarian neoplasms or intraabdominal tumours.[1,7] Leiomyomas usually cause symptoms of heavy menstrual period, infertility, abdominal distension and pressure symptoms that may result in sensation of bowel distension, increased urinary frequency and respiratory difficulty.[3,4]

They are easily recognized on imaging except with atypical presentation caused by degenerative changes.[1] Definitive diagnosis was by histology that showed proliferation of smooth muscle cells without atypia.[1,3] Surgery is the treatment of choice for giant uterine leiomyoma.[4] Presented is a 28-year-old with progressive abdominal swelling over 9 months duration. She was seen at the surgical outpatient department on account of aesthetic displeasure that arose from an intraabdominal swelling without pregnancy. Following surgery, a definitive diagnosis of multiple leiomyoma with a large subserous uterine leiomyoma with extensive cystic degeneration. We present a 28-year old woman with a large subserous uterine leiomyoma that presented as an intraabdominal tumor to highlight the diagnostic challenges that may arise.

Case report

A 28-year-old woman presented to our facility in October 2023 with abdominal swelling in the past 9 months. It was insidious in onset, first noticed from her right flank and it progressively increased in size extending to the upper abdomen more on the right extending to the left. Swelling was painless and there was no swelling elsewhere in her body. There was no history of anorexia, vomiting, easy satiety, change in bowel habit, passage of dark tarry/blood in stool nor weight loss. Furthermore, there was no history of jaundice, urinary symptoms nor family history of similar swelling. Patient had dyspeptic symptoms for about 5 years. She had appendicectomy 12 years ago. She's Para 0 + ^{2} (termination of pregnancies), noticed some episodes of scanty periods and shortened period of her menstrual flow (2 days). Her LMP was mid-August 2023.

Examination showed a young woman otherwise healthy looking, not pale, afebrile, anicteric and no pedal oedema. Chest was clinically clear. The abdomen was asymmetrically distended with a swelling that was estimated to have a fundal height of 32 weeks in the right half of the abdomen and extended across the midline, Figure 1(a) and (b). There was an oblique scar in the right iliac fossa from appendectomy Figure 1(a). Swelling was spherical, measured 30 cm × 25 cm. It was non-tender, was possible to get above and below the swelling that was firm, smooth, mobile and not attached to overlying skin. There was no demonstrable ascites and rectal examination was unremarkable. A clinical diagnosis of an intraabdominal mass was made.

Work up included full blood count (FBC), urea, electrolytes and creatine (EUCr), clotting profile that were unremarkable. Urinalysis was normal. Abdominopelvic CT scan presented in the clinic showed a huge loculated cystic hypodense intrabdominal mass with thick enhancing wall occupying predominantly the right flank and measured 18.4 cm, Figure 2(a). It displaced the abdominal aorta and IVC to the left. Uterus was reported as bulky with AP dimension of 8 cm showing a calcified myoma (5.0 × 5.5 cm) in its myometrium that compressed the adjacent bladder, Figure 2(b). The impression was: intra-abdominal mass with calcified uterine fibroid. There was no gynecological consultation in the preoperative period.

During operative laparotomy the gynecologist was invited with the following findings:

1. Large subserosal pedunculated spherical uterine mass, FIGO type 7 that measured about 25 cm in the widest dimention that arose from the right side of the fundus of the uterus, Figure 3(a) and (b).

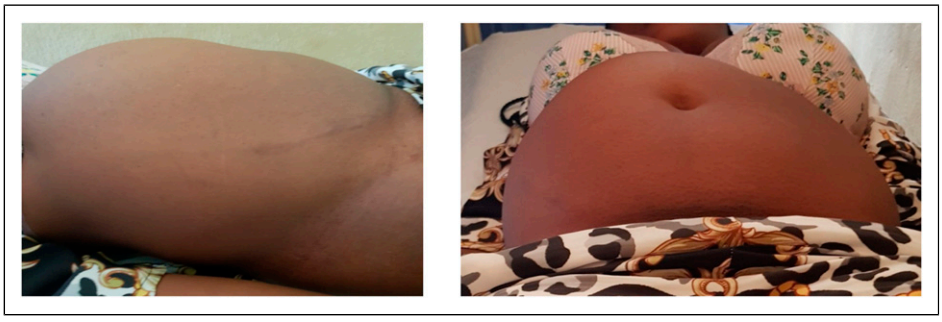

Figure 1. a, b- Clinical photographs showing abdominal distension, (a)- right lateral view in addition showed a post appendectomy scar on the right lower abdomen. (b), from the foot of the bed showing asymmetrical distension, more on the right side.

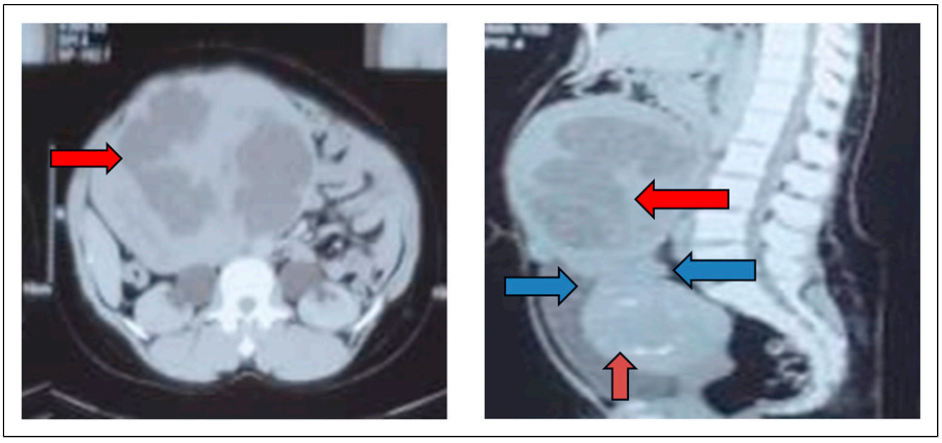

Figure 2. a, b- CT- Axial section showing intraabdominal mass with degenerative changes (red arrow), (b)-CT- Sagittal section showing abdominopelvic mass with "ultrashort pedicle sign" (blue arrows), dystrophic calcification of cervical myoma (brown arrow) and cystic degenerative changes (red arrow).

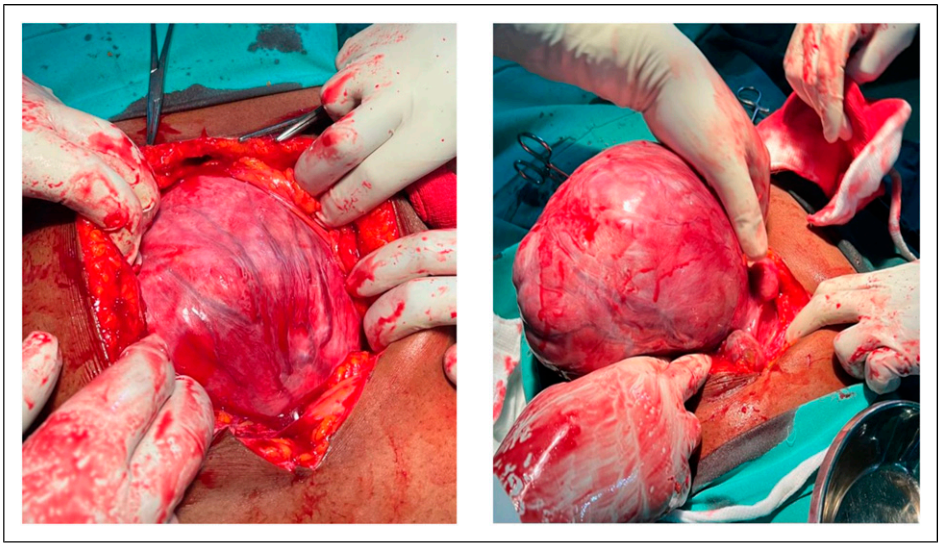

Figure 3. (a)- Large subserous uterine leiomyoma insitu, (b)- Intraoperative, demonstrating the ultra-short pedicle.

2. Spherical subserosal uterine mass (that measured 3.5 cm in the widest dimention) attached to the supero-anterior aspect of the uterus, Figure 4(a).
3. Intra-uterine mass located at the anterior part of the fundus, Figure 4(b).
4. Large spherical intra-cervical mass that measured about 10 cm in its widest dimention, Figure 4(b)
5. Normal adnexa, liver, spleen, mesentery and no ascites.

Patient in supine position, under general anaesthesia with endotracheal intubation, nasogastric tube was passed to deflate the stomach, a urethral catheter inserted to empty the urinary bladder/monitor hourly urinary output. The abdomen was accessed through a midline incision extending from the xiphisternum to the suprapubic area with hemostatic control. A huge mass was delivered that revealed a large subserous uterine mass with an ultra-short peduncle attached to the right side of the uterine fundus, FIGO 7, Figure 3(a) and (b). Another relatively smaller subserous masses was also noted, Figure 4(a). No adhesions were noted. The serosa was deroofed at the peduncle and the mass was excised between two myometrial clamps. The myometrial tissue was transfixed with polyglactin size two and the serosal cover restored. The Gynecologist who was invited discovered and removed another uterine mass located at the cervix, Figure 4(b), alongside another subserous mass, 4a. There was associated focal adenomyosis in the left fundal area also excised, (Figure 4(b)-part held between the

thumb and fingers). Mass closure was with polyamide two monofilament. Skin was apposed with polyamide 2/0 interrupted vertical mattress sutures and the harvested specimens sent for histology, Figure 5(a) and (b). Estimated blood loss was 350 mls. Post operative period was uneventful and patient discharged.

Histology was reported as follows: macroscopy-four different specimens, largest pedunculated subserous, cervical, left fundal and subserous masses that measured 18 × 17 × 13 cm, 11 × 9 × 5 cm, 3.5 × 3 × 2 cm and 3.5 × 2.5 × 0.5 cm respectively. Cut surface of the largest mass (subserous uterine mass) showed multiple cystic spaces, the largest measured 10.5 cm and smallest 0.5 cm across the widest dimensions, Figure 6. Microscopy revealed: uterine leiomyoma that showed bundles of smooth muscle fibers that were haphazardly arranged. Individual muscle fibers were spindle shaped with bipolar eosinophilic cytoplasm with cigar shaped nuclei. There were areas that showed hyalinization while the other areas showed cystic spaces and there was no atypia. Furthermore, this uterine leiomyoma showed degenerative changes (hyaline and cystic), Figure 7(a) and (b). Histology of the subserosal and cervical intrauterine masses showed, interlacing bundles of smooth muscles fibers that were haphazardly arranged, individual

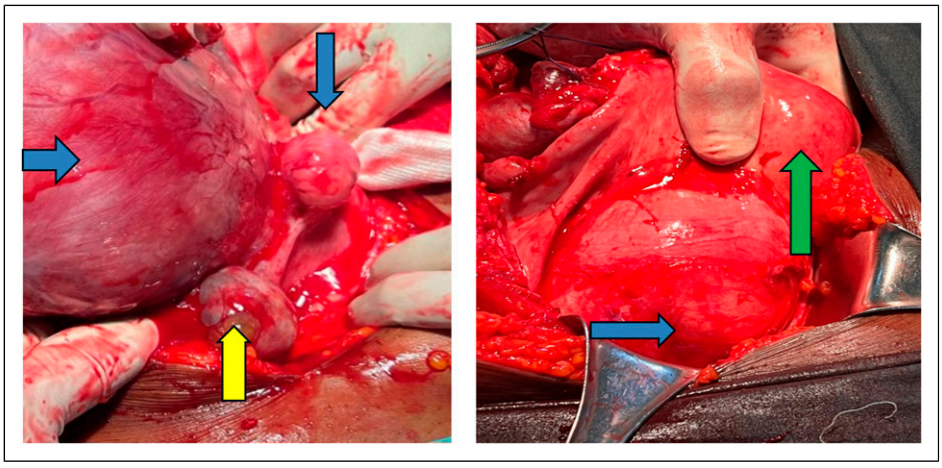

Figure 4. (a)- Multiple subserous leiomyomas (blue arrows), right ovary (yellow arrow), (b)- cervical leiomyoma (blue arrow) and adenomyosis (green arrow).

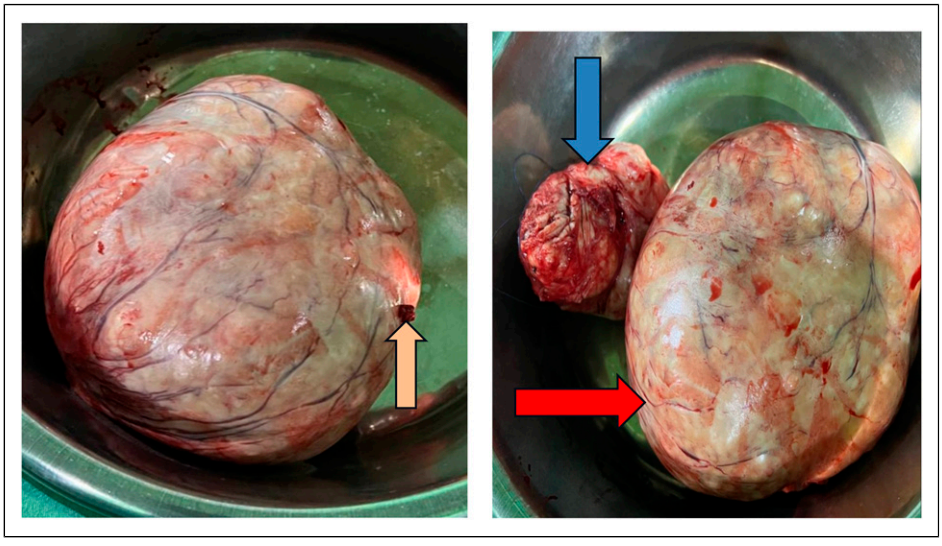

Figure 5. (a)- Gross picture of subserous uterine leiomyoma- site the attachment of the pedicle (green arrow). (b)- Gross picture of subserous uterine leiomyoma (red arrow), Leiomyoma of the uterine cervix (blue arrow).

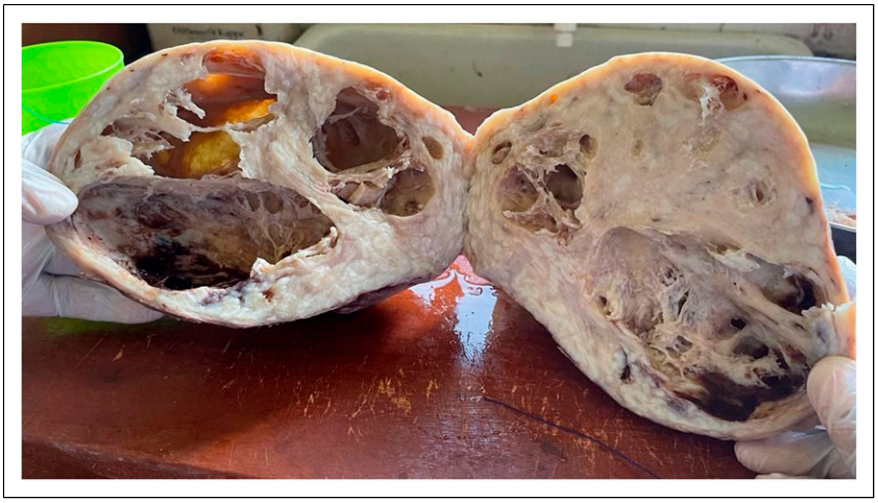

Figure 6. Cut surface of intraabdominal subserous uterine leiomyoma, showing multiple cystic spaces, largest 10.5 cm in the widest dimension and the smallest 0.5 cm.

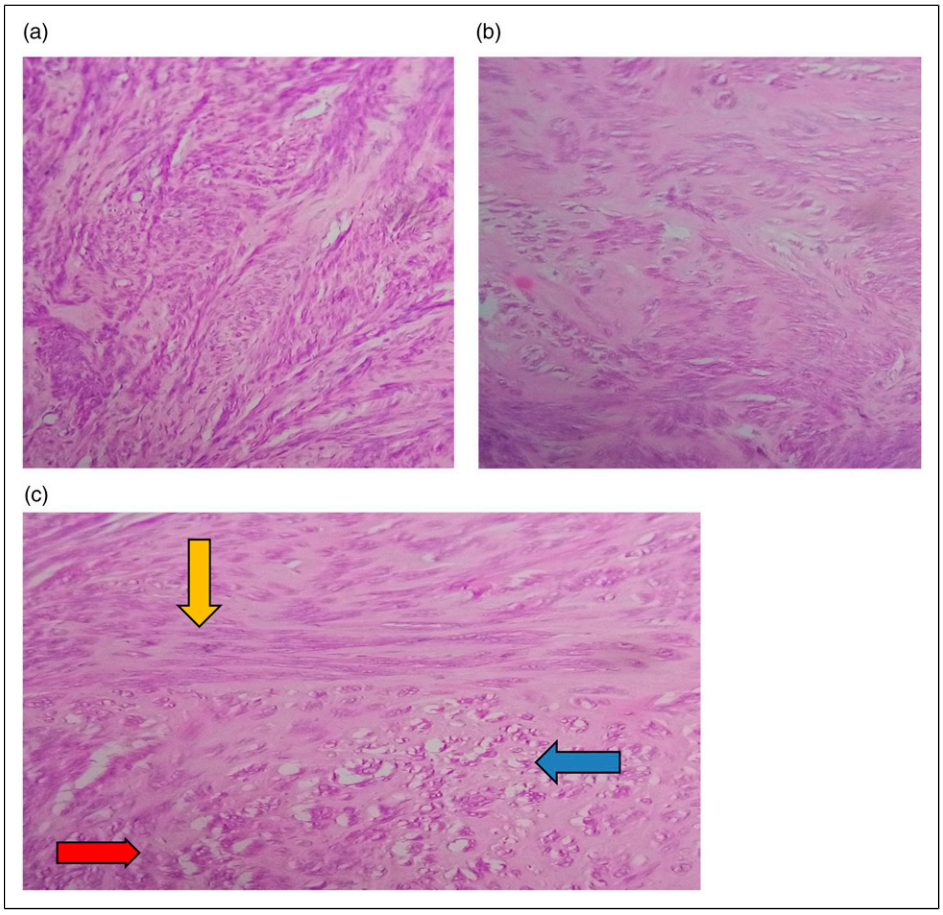

Figure 7. a, b- Uterine leiomyoma. (a)- Histology of intraabdominal subserous uterine leiomyoma showed bundles of smooth muscle fibers that were haphazardly arranged, individual muscle fibers were spindle shaped with bipolar eosinophilic cytoplasm with cigar shaped nuclei. (b)- showed hyaline degeneration. There was no atypia, (c)- Uterine adenomyosis, blue arrow-endometrial glands, red arrow-endometrial stroma, yellow arrow-myometrium.

muscle fibers were spindle shaped with bipolar eosinophilic cytoplasm with cigar shaped nuclei with no atypia-subserous and uterine leiomyomata. Left intrauterine fundal mass showed, sections of the endometrial glands and stroma deep in the myometrium of smooth muscle fibers that were haphazardly arranged. The individual muscle fibers were spindle shaped with bipolar eosinophilic cytoplasm with cigar shaped nuclei-uterine adenomyosis, Figure 7(c). The definitive diagnoses were multiple uterine leiomyomas and adenomyosis. She was referred to the gynaecologist for further care.

Discussion`

Leiomyomata of the uterus are gynecological problems in women of reproductive age with negative impact on their health.[3] They are the most common form of benign tumors of the female reproductive tract.[1] However, giant uterine leiomyoma are rare with about 100 cases documented worldwide.[3] Pedunculated subserous uterine tumor in the abdominal location can attain a giant size with diagnostic and surgical challenges especially with nonspecific symptoms as was the experience with our patient. She presented to the surgical outpatient mainly on the account of the aesthetic discomfort occasioned by the increasing abdominal girth in a non-pregnant state. Uterine leiomyoma is more common in nulliparous women and prevalent in black women compared with the Caucasians, Asians or Hispanic women.[3] It occurred in 20%–30% of females older than 30 years. At the age of 50 years, 80% of African and about 70% of Caucasian women had uterine leiomyoma.[2] In another study, it was estimated that by the 5th decade, as many as 50% of women of African descent had leiomyoma.[3]

The etiology of uterine leiomyoma remain unknown with several risk factors identified: positive family history, ovarian hormones, genetic mutations and lifestyle related factors. The later included diet, obesity, medical contraception, smoking and exercise. The major hormones implicated in the growth of leiomyoma were ovarian steroids (estradiol and progesterone).[2] The population of hormone receptors have been known to be increased in fibroids.[3] The incidence of fibroid is decreased with prolonged use of oral contraceptive pill as well as increased number of term pregnancies.[3] The growth of leiomyoma appeared to be dependent on the hormone estrogen. If a woman with leiomyoma is still menstruating, the tumour will probably continue to grow slowly.[7] Progesterone down-regulates apoptosis in the tumor while estrogen increase production of extracellular matrix.[3] Cytogenetic abnormalities occur in 50% of fibroids, most commonly, translocation within or deletion of chromosome, translocation of chromosome 12 and 14 and occasionally structural aberrations of chromosomes.[3] MED 12 gene is associated in the pathogenesis of leiomyoma. MED 12 negative genotype is associated with larger fibroids.[5] Mutations in the gene encoding fumarase hydratase were shown to predispose women to multiple leiomyomas.[3]

The position of the leiomyoma in relation to the uterus determined the patients symptoms and diagnostic specificity.[2] The most common location is the body of the uterus and may also involve the uterine cervix in minority of instances,[1] our patient presented with multiple leiomyomas of the corpus and cervix. Based on their positions within the uterine wall, leiomyomas were classified as: (a) intramural (70%), (b) uterine cavity (10%)- submucosal, pedunculated submucosal or pedunculated vaginal, (c) growing outwards from the uterus (20%), further classified as cervical, subserous, intraligamentous or pedunculated subserous (abdominal).[8] In a similar classification, it was divided into extra-uterine and uterine fibroids which is further classified into intramural/interstitial (75%), submucosal (5%), subserosal (10%) and cervical (1%).[5] In the submucosal (uterine cavity) or subserosal (abdominal) they may have a connecting stalk (pedunculation).[2] The above locations determined the pattern of clinical presentation. Clinical presentation in majority of cases may be asymptomatic. The symptomatic women were most likely to present with uterine bleeding: heavy menstrual period and frequent menstruation.[2] In addition, frequent symptoms include dyspareunia or chronic acyclic pain, they can affect fertility with remarkable psychological impact on a woman's life.[2] Our patient had amenorrhea of about 2 months, this should have prompted a pregnancy test.

Uterine leiomyoma has been described as giant based on the weight of 11.4 kg or greater or have a diameter greater than 17 cm or dimension of 33 × 28 ×22 cm.[9] Due to the ability of the anterior abdominal wall become distended and the large volume of the abdominal cavity, uterine leiomyoma (especially the subserous pedunculated) can grow into extremely large dimensions.[3] This can result in a feeling of dragging sensation of the abdomen resulting in aesthetic displeasure from increased abdominal girth in keeping with the experience of our patient. As the tumor grows, myomas can result in complications from compression related symptoms of dyspnea, frequent urination, bowel complaints[2,3] and other unpredictable events as massive bleeding, adhesions to surrounding organs including displacement of organs/adjacent structures.[4] The index patient demonstrated hydroureters as reported in the CT scan, the patient was however asymptomatic, reported also, was the displacement of the descending aorta and the inferior vena cava (IVC). Kim et al reported a pedunculated leiomyoma in an umbilical hernia in a non-pregnant woman with a large intra-abdominal lesion.[10] Pedunculated subserous myomas may result in an emergency arising from the complication of torsion.[2] Leiomyoma may occur with endometriosis and adenomyosis with overlapping symptoms.[2]

As uterine leiomyoma outgrow their blood supply, complications may arise resulting in degeneration: hyaline, red, myxoid or cystic changes and dystrophic calcification.[1,5] Rarely uterine leiomyoma may undergo malignant degeneration resulting in sarcoma in less than 1.0% with some reports estimated to be as low as 0.2%.[1,2,5] Uterine leiomyomas have been misdiagnosed as adenomyosis, hematometra, uterine sarcoma, ovarian masses, pregnancy and gastrointestinal tumours especially gastrointestinal stomal tumours (GIST).[1,2,11]

Preoperative imaging studies are useful to determine the extent of the tumor and ultrasound sonography (USS) is the preferred modality of initial evaluation as it is easily available, non-invasive and convenient cost-benefit ratio. An USS especially vaginal one can help to assess the mass, vascularity, pedicle, adenxa, ascites and kidneys.[1,2] This was a significant diagnostic deficiency in our diagnostic work up. Uterine leiomyomas are often detected on CT and may be incidental. Uterine leiomyoma and normal myometrium are sometimes not distinguishable on CT except when they are calcific or necrotic changes.[12] The CT of the patient revealed a pedicle sign, cystic degenerative signs may create heterogenous or an unusual appearance that contributed to the diagnostic challenges.[1,11] Atypical appearance of leiomyoma limits its preoperative informative value of some of the imaging modalities[2] as was demonstrated in our patient with extensive degenerative changes. MRI is more specific in the determination of the origin of uterine leiomyoma including degenerative changes. The multiplanar imaging may reveal a vascular pedicle or another form of attachment of uterine leiomyoma. Non-degenerated leiomyoma shows characteristic low-intermediate signal intensity on T1-W1 and low signal intensity on T2-W1. Degenerated leiomyomas are variable in terms of MRI signal characteristics. Myxoid degeneration and necrosis may present as high signal intensity areas on T2-W1 without enhancement. Cobblestone-like tissue attributable to hyaline degeneration with foci of high signal intensity that represented areas of infarction caused by rapid growth seen on both T1 and T2-W1. No imaging modality can exclude malignancy leaving the diagnosis of a giant uterine fibroid a challenge.[2] Histologically, smooth muscle cells proliferate, occasionally histologic composition may not allow precise separation from intestinal organs.[2] Diagnostic laparoscopy is valuable especially in the resolution of diagnostic challenges and in some instances a preferred route for surgery.[5]

Treatment is individualized based on the severity of symptoms and the need to preserve fertility.[12] Surgical options include myomectomy and abdominal hysterectomy, the main indication for myomectomy is the preservation of the uterus for childbearing in the younger-aged woman as was indicated in our patient.[3] Robotic-assisted laparoscopic myomectomy (RALM) is the treatment of choice and gold standard.[13] Abdominal hysterectomy is, however the most effective surgical therapy for giant uterine leiomyoma except when pedunculated or when fertility is an issue and this should be discussed with the patient.[3] Giant uterine leiomyoma cannot be treated with minimally invasive surgery.[2] Complications of surgery include massive blood loss and others like injury to adjoining structures occasioned by adhesions/displacement to surrounding organs and infection.[4,12] Jonas et al reported a perioperative mortality of 14.8%–16.7% among patients with giant uterine leiomyoma.[4] Follow up of patients is aimed at the detection of recurrence. Follow up of patients is for the detection of the recurrence of myomas and this will commence within 2–6 weeks after surgery. Ultrasonography and pelvic examination are commenced. Subsequent visits are scheduled for 3 months, 6 months and 1 year. If there is no recurrence in a year, annual examinations are likely to be adequate. However, no studies have been done in support of this protocol.[14] Prevention of massive uterine leiomyoma requires close surveillance and early surgical treatment.[2] Treatment of lifestyle-associated risk factors and vitamin D supplementation, use of statin and dietary modifications appears to be productive along with parity.[2]

Conclusion

In conclusion, large subserous uterine leiomyoma should be considered in a patient of childbearing age with a grossly distended abdomen without obvious evidence of pregnancy or malignancy. Detecting the continuity of an abdominal mass (even with extensive cystic degenerative changes) with the uterus by a stalk (pedicle sign) on imaging in the absence of ascites should arouse the diagnosis of pedunculated subserosal leiomyoma. Large subserous uterine leiomyoma may present a diagnostic and surgical challenge that require expertise and interdisciplinary cooperation. It can be successfully managed without complications with proper diagnosis and surgical expertise.

Acknowledgements

We acknowledge the patient and all the participating authors for their contribution to the paper.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

This research received no financial support for the research, authorship, and/or publication of this article.

Contributorship

Dr Ashindoitiang for concept to write the article and drafting of the manuscript, Dr Nwagbara participate in writing the manuscript Dr Effiong Edet participate in gathering materials Dr Ugbem for histological diagnosis, Dr Ukam for analysis and Prof Asuquo for surpervising the writing of the manuscript. All authors contributed to different aspect of the report.

Ethical statement

Ethical approval

The requirement for ethics approval was waived because this study was a case report of a patient managed by our team, not an interventional study (i.e., no intervention or experimentation was carried out on the patient for the purpose of this study).

Informed consent

The patient provided both verbal and written informed consent for the reporting of this case and all accompanying images. Consent granted by the patient to use clinical pictures.

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