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Case Report

Atypical leiomyoma: unusual locations and findings coupled with uncommon presentation

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ABSTRACT

Extrauterine leiomyomas are a rare phenomenon, and they present a great diagnostic challenge. Retroperitoneal leiomyomas remain asymptomatic for a variable period and usually present with wide variety of signs and symptoms. A 51 years old female was multiparous and had undergone a total laparoscopic hysterectomy along with a bilateral salpingo-oophorectomy about four years ago. She visited the outpatient department of a tertiary care hospital complaining of something coming out per vaginum and expressed difficulty in passing urine and also complained of stress urinary incontinence. Bimanual pelvic examination, ultrasonography (USG) and Magnetic resonance imaging (MRI) revealed a large oval shaped well defined solid mass completely filling the vaginal canal while compressing urinary bladder. Tumor marker levels were normal. Laparotomy with adhesiolysis and complete excision of retroperitoneal mass was performed. Frozen section followed by histopathology and immunohistochemistry confirmed that the mass was a leiomyoma. After surgical intervention and removal of mass, the symptoms dissipated and patient had a complete recovery. Despite hysterectomy the manifestation of leiomyoma appears to be apparent. Surgical excision of the mass appeared to be the correct move forward after the leiomyoma was diagnosed with the help of the MRI and the USG.

Keywords: Retroperitoneal, Leiomyoma, Laparotomy, Hysterectomy, Immunohistochemistry

INTRODUCTION

Uterine leiomyoma that are manifested clinically affect 20%–30% of women above the age of 35 years.[1,2] Extra-uterine leiomyoma is an uncommon phenomena, and they present a great diagnostic challenge. Retroperitoneal leiomyomas, defined as those located posterior to parietal peritoneum, are a very rare occurrence.[1-4] As of 2007 (1941-2007), only 37 retroperitoneal leiomyomas have been reported in the literature with comprehensive information. There is a need for further research on this condition. Poliquin et al studied 100 cases of retroperitoneal masses and, of these, only one showed a direct connection with the uterus at the level of the internal cervical os.[5]

More than 40% of patients affected by a retroperitoneal leiomyoma have a concurrent uterine leiomyoma or a history of undergoing a hysterectomy for treatment of a uterine leiomyoma.[5] However, some extra-uterine leiomyomas may mimic malignancies, and serious diagnostic errors may result. The most useful modalities for detecting extrauterine leiomyomas are ultrasonography (USG), computed tomography (CT), and magnetic resonance (MRI) imaging.[2] Rarely, the extrauterine masses may extend to the upper retroperitoneum, as high as the level of the renal hilum. They may enlarge considerably yet remain asymptomatic and be detected incidentally at a routine check-up or autopsy. Common symptoms include discomfort, fatigue, and back pain.[6] The lesions are hormonally responsive

and have clinical manifestations similar to those of uterine leiomyomas.

This case report describes retroperitoneal leiomyoma presented with something coming out per vaginum and difficulty in passing urine with stress urinary incontinence.

CASE REPORT

A 51 years old woman of Indian ethnicity approached the out-patient department in a tertiary care hospital located in Mumbai, India with complaints of something coming out of vagina since 3 months. She complained that this mass had increased gradually in size. She also complained that for the past six months, she had a difficulty in initiating micturition and had observed dribbling of urine during coughing, laughing or lifting weights. This symptom had also worsened in the last three months.

She had undergone total laparoscopic hysterectomy and bilateral salpingo-oophorectomy 4 years ago for refractory menorrhagia with fibroid uterus which was confirmed on histopathological examination. She has had two children via normal vaginal delivery. No other surgical or medical history was available from the patient.

On physical examination, the patient appeared to show normal findings with no palpable masses felt abdominally. Pelvic examination revealed an oval shaped mass displacing posterior wall of vagina and filling vaginal orifice completely. Bimanual pelvic examination revealed a large solid mass of firm consistency in the midline just above the vault. The mass had restricted mobility.

A pelvic ultrasound was arranged and a blood test to check for serum tumor markers (CA 125, CA 19.9 and CEA) was conducted. All tumor markers were within normal limits. The pelvic ultrasound showed a solid pelvic mass arising just distal to the site of surgical stump measuring 5.6 x 5.5 x 5 cm (LxHxB). There was suspicious of a fibroma or a leiomyoma (Figure 1). There was no evidence of hydronephrosis in either kidneys and no ascites was observed. Due to uncertainty of the origin of the mass, a MRI of the pelvis and abdomen was conducted. It showed a large, oval shaped solid pelvic mass of 7.6 x 4.3 x4.4 cm (LxHxB) size. The mass was filling the vagina and compressing urinary bladder from posterior aspect. (Figure 2) There was no invasion of bladder or rectum. No definite organ of origin could be identified. There was no intraabdominal lymph node enlargement. The kidneys, liver, spleen, pancreas and adrenal glands appeared to be normal.

Due to the past history of the total abdominal hysterectomy and bilateral salpingo-oophorectomy along with the uncertainty of the origin of the mass, the patient was scheduled for laparotomy. The findings from the laparotomy revealed a pelvic mass of 7.5 x 4.5 x 2.5 cm (LxBxH) size posterior to urinary bladder, which was adherent to the colon and vaginal wall. The adhesions were separated from the mass, and the mass was completely removed and sent for frozen section. Frozen section report was suggestive of benign spindle cell neoplasm favouring leiomyoma. Histopathology was suggestive of benign spindle cell neoplasm favouring leiomyoma with no evidence of necrosis/nuclear atypia. Immunohistochemistry studies confirmed the mass as leiomyoma. (Desmin- positive, Caldesmon - positive, SMA- Patchy positive, Ki 67 <2%)

The patient was discharged on the 5th post-operative day. The patient had a complete recovery. At the follow-up appointment, a month later, she remained symptom free.

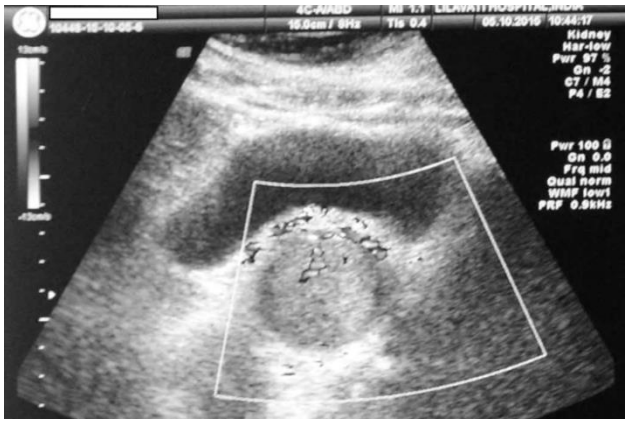

Figure 1: Pelvic USG showing an oval shaped solid pelvic mass arising just distal to the site of surgical stump measuring 5.6 × 5.5 × 5 cm compressing urinary bladder.

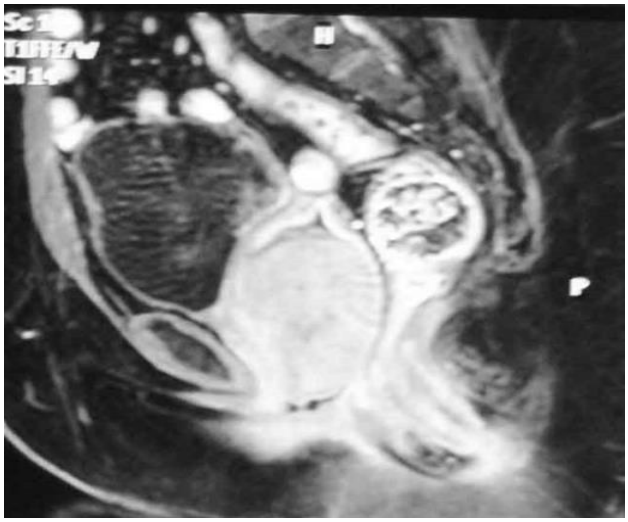

Figure 2: MRI (abdo + pelvis) showed large, oval shaped soft tissue mass in the pelvis of 7.6 × 4.3 × 4.4 cm size filling the vagina and compressing urinary bladder from posterior aspect. No bladder/rectum invasion. No lymphadenopathy.

DISCUSSION

Uterine fibroids are the most common benign solid pelvic tumors in women and are present in about 80% of all hysterectomy specimens. Retroperitoneal leiomyoma has a rare occurrence and has recently been recognized as distinctive lesions with similar histological features as uterine leiomyoma.[7] This case report outlines the significance of considering solitary fibrous tumors as a differential diagnosis for a pelvic mass.[5,6,8,9]

Solitary fibrous tumors (SFT), or commonly known as fibromas, were first documented at the turn of the twentieth century, originating in the intrathoracic cavity, typically from the pleura. Fibromas are slow growing, well circumscribed, spindle cell neoplasms arising from the mesenchyme. These histologically benign tumors, which originate from smooth muscle cells, usually arise in the genitourinary tract (in the vulva, ovaries, urethra, and urinary bladder) but may arise in nearly any anatomic site. In addition, unusual growth patterns may be seen, including benign metastasizing leiomyoma, disseminated peritoneal leiomyomatosis, intravenous leiomyomatosis, parasitic leiomyoma, and retroperitoneal growth.[2,6]

More than 40% of patients affected by this retroperitoneal condition have a concurrent uterine leiomyoma or a remote history of hysterectomy for treatment of a uterine leiomyoma.[5] However, some extrauterine leiomyomas may mimic malignancies, and serious diagnostic errors may result. With regard to their pathologic origin, it is unclear whether these retroperitoneal lesions represent metastatic or synchronous primary lesions and whether they arise from the hormonally sensitive smooth muscle elements or from the embryonal remnants of müllerian or wolffian ducts. Zaitoon suggested the parasitic theory for such tumor growth, while Stutterecker et al claimed that müllerian cell rests or smooth muscle cells in the retroperitoneal vessels wall are the putative origin.[10,11] Kho and Nezhat proposed an 'iatrogenic' origin for such growths mostly associated with previous abdominal operations (83%) and myomectomies (67%), most of them via laparoscopy with morcellation.[12] Our case also supports iatrogenic origin of retroperitoneal leiomyoma.

They may enlarge considerably yet remain asymptomatic and be detected incidentally at a routine check-up or autopsy. Common symptoms include discomfort, fatigue, and back pain.[1,3,6,8,9] Some present with dysmenorrhea and dyspareunia.[4,13] While some present with urinary and bowel complaints.[1] The lesions are hormonally responsive and have clinical manifestations similar to those of uterine leiomyomas.[14]

The most useful modalities for detecting extrauterine leiomyomas are ultrasonography, computed tomography, and magnetic resonance (MR) imaging.[2] Rarely, the extrauterine masses may extend to the upper retroperitoneum, as high as the level of the renal hilum.[4] USG may depict a well-defined mass with a variable but usually homogeneous echo texture within the retroperitoneum. Homogeneous attenuation is typically seen within the mass at CT. Smooth muscle tumors within the retroperitoneum are usually malignant. It is not possible to differentiate leiomyoma from leiomyosarcoma on the basis of imaging features alone, although extensive central necrosis, invasive growth, and a heterogeneous appearance are suggestive of leiomyosarcoma.[2]

The differential diagnosis of a pelvic fibroma most commonly includes leiomyoma or leiomyosarcoma and fibrosarcoma.[6,15] Other differential diagnoses include common benign and malignant neoplasms of neurogenic origin (schwannoma, paraganglioma, ganglioneuroma, extraadrenalpheochromocytoma) as well as teratoma, desmoid tumor, hemangioma, extra-adrenal angiomyelolipoma, sarcoma, lymphoma, and metastatic tumors.

Surgical removal of the mass is the main stay of treatment, which can be by laparotomy or laparoscopic removal.[1,4,6,16] Isolated retroperitoneal leiomyomas and pelvic retroperitoneal leiomyomas are rare occurrences, and the prognosis for patients with these lesions is good.[4-6,14]

CONCLUSION

Retroperitoneal leiomyomata is a rare occurrence. Through imaging techniques especially USG, CT or MRI one can obtain help in operative planning. However, these techniques may also miss the exact site and size of these fibroids. Final diagnosis is made intraoperatively on direct visualisation. Complete excision with or without abdominal hysterectomy is the treatment of retroperitoneal fibroids. In case of isolated retroperitoneal leiomyomas and pelvic retroperitoneal leiomyomas, the prognosis is good.

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