

Angioleiomyoma Uterus — A Rare Tumor

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Introduction

Angioleiomyoma (AL) is a benign tumor of mesenchymal origin containing thick walled vessels. It is more common in middle aged women.¹ It typically occurs as a solitary subcutaneous painful nodule and is more common in the lower extremities.² The uterus is extremely rare site of Angioleiomyoma. The etiopathogenesis of uterine Angioleiomyoma is not well established.³

Case Report

A 37-year-old nulliparous woman presented to the OPD of IMS and SUM Hospital with complaints of heaviness and gradual swelling of abdomen over a period of two months duration along with history of heavy menstrual bleeding of six months duration. Her cycles were of 35 days duration associated with heavy menstrual flow and mild abdominal discomfort. She also complained of incomplete evacuation of bladder and increased frequency of micturition over the last 3 months. She had normal bowel habits, without

any comorbidities. She hails from a family of middle socio-economic strata with no history of any kind of addiction.

During physical examination she was found to be average built with a BMI of 23 kg/m². She was pale without icterus, clubbing, and lymphadenopathy. No thyromegaly was observed and her breast examination revealed no abnormalities. Respiratory and CVS examinations revealed no obvious clinical abnormality and were essentially normal. Her abdominal examination revealed a mid-line, nontender, cystic mass of 36 weeks size with smooth surface, regular margin and the mass appeared to be originating from the pelvis. On percussion there was a dull note over the mass and shifting dullness was absent and on auscultation soufflé was heard. Pelvic examination revealed the mass to be continuous with the uterus. Both the fornices were filled with the mass. Ultrasonography showed a huge abdomino-pelvic cystic mass lesion of size (22x30cms) with eccentric wall thickening and increased vascularity. (RI 0.43) Ovaries could not be visualised separately. Ovarian Tumor markers HE 4, CA 125 and ROMA score were elevated. CECT revealed (Fig 1) a bulky uterus with multiple myomas and adenomyotic changes with large complex solid cystic lesion (21.2 X 29.4 X 31.8 cms) arising from anterior uterine wall with features of extension. Suggested possible diagnosis were (a) Adenomyosis with large Endometriotic Cyst or (b) Sarcomatous transformation of a large anterior wall uterine myoma.

Laparotomy was planned in view of Uterine Sarcoma/Ovarian tumor. Case along with the prognosis was explained to the patient and her family members in

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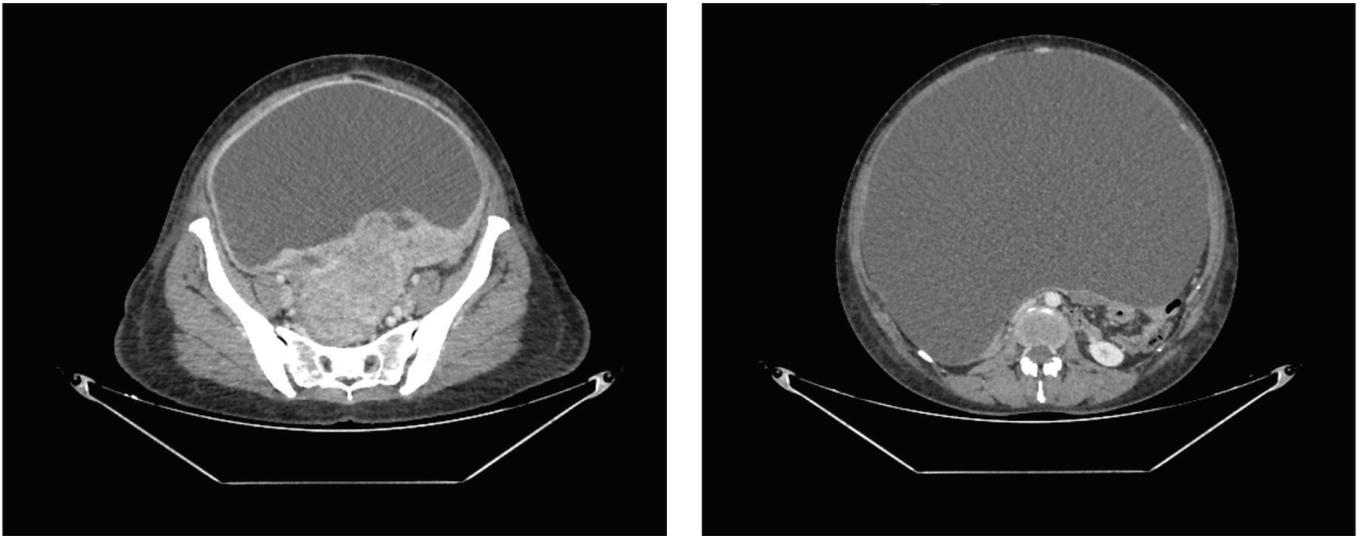


Fig.1: Contrast enhanced computed tomography (CECT) showing large complex solid cystic mass

both situations. Consent obtained for Hysterectomy and pelvic lymph node sampling. On laparotomy a huge cystic mass with hemorrhagic fluid was detected after opening the peritoneum of extended midline vertical incision. Mass found to be arising from the uterus. Peritoneal cavity was devoid of free fluid. Approx 10 litres of hemorrhagic fluid suctioned out of the mass (Fig-2). Total abdominal hysterectomy with bilateral salpingo-oophorectomy was done. All abdominal organs along with omentum and paracolic gutter were explored for any tumor deposit or any abnormality which was not detected. All tissue and aspirated fluid were sent for histopathological study.

On gross examination, the uterus was asymmetrically enlarged with multiple intramural fibroids noted with the largest measuring 3.5x2.5x2 cms. A large solid cystic mass of size 30x17x7cms arising from the

fundus of the uterus was noted. The outer surface was firm and whitish with areas of congestion. The cut surface revealed whitish to tan appearance with no hemorrhagic or necrotic areas in between. The Histopathological examination revealed angioleiomyoma (Vascular Leiomyoma) of the uterus (Fig-3).

Post-operative period was uneventful and the patient was discharged on the seventh postoperative day with advice of regular follow-up.

Discussion

Angioleiomyoma is also known as Vascular leiomyoma. Angioleiomyoma is classified into 3 types histologically: Capillary or solid, Cavernous and venous. The index case of AL of Uterus was first reported in 2003 by Hsieh et al. She had menorrhagia

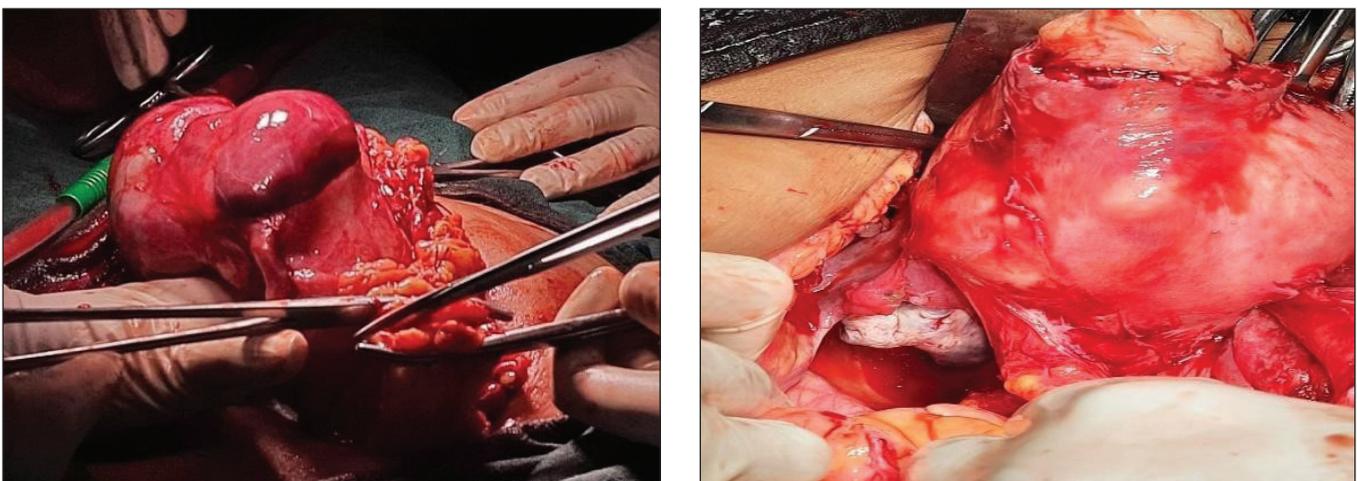


Fig. 2: Gross photographs showing a large mass arising from the uterus

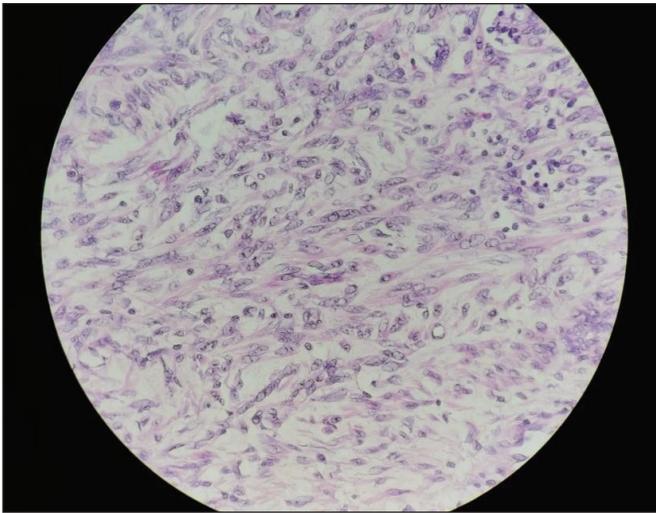


Fig. 3: Histopathological findings showing tumour cells arranged in short and dense fascicles whorled with intervening myxoid areas. Many moderate to small sized vessels are seen within the tumour.

and severe anaemia.⁵ Our case was also presented with menorrhagia, abdominal mass and pain in the abdomen. Menorrhagia can possibly be because of dysregulation of different growth factors and/or because of receptors, which regulate the process of

angiogenesis. Pain may be attributed to intratumoral hemorrhage.

In previously reported cases, patients have presented with symptoms of abnormal uterine bleeding,^{4,5} palpable abdominal mass,⁵ pain in abdomen⁶ and pressure symptoms.⁷

As proposed by Hsieh et al, typical CT findings of AL are a multilobulated mass with solid and laminated configuration.⁵

The treatment of ALs both uterine and extra uterine, is complete surgical excision of the tumor. In cases involving the uterus either Hysterectomy or excision of the tumor may be performed. Outcome of both the treatment modalities are good.

Conclusion

Pre-operative diagnosis of AL is rarely possible. It is usually diagnosed by histopathology. Though extremely rare AL should be kept in Differential Diagnosis of Uterine or adnexal mass. As suggested by Sahu et al² they should be included in WHO classification of tumors of female genital tract as a benign leiomyoma variant.⁸

REFERENCE

- Garg G, Mohanty SK. Uterine angioleiomyoma: a rare variant of uterine leiomyoma. Arch Pathol Lab Med. 2014 Aug; 138(8): 1115-8. doi: 10.5858/arpa.2013-0315-RS. PMID: 25076303.
- Sharma C, Sharma M, Chander B, Soni A, Soni PK. Angioleiomyoma uterus in an adolescent girl: a highly unusual presentation. J Pediatr Adolesc Gynecol. 2014 Jun; 27(3): e69-71. doi: 10.1016/j.jpag.2013.07.002. Epub 2013 Oct 9. PMID: 24119657.
- Singh S, Naik M, Bag ND, Patra S. Angioleiomyoma of Uterus Masquerading as Malignant Ovarian Tumor. J Midlife Health. 2017 Jul-Sep; 8(3): 145-147. doi: 10.4103/jmh.JMH_47_17. PMID: 28983163; PMCID: PMC5625580.
- Agorastos T, Dinas K, Patsiaoura K. Cystic degenerated angioleiomyoma mimicking ovarian pathology. Acta Obstet Gynecol Scand. 2001 Sep; 80(9): 863-5. doi: 10.1034/j.1600-0412.2001.080009863.x. PMID: 11531639.
- Hsieh CH, Lui CC, Huang SC, Ou YC, Chang Chien CC, Lan KC, Huang KH. Multiple uterine angioleiomyomas in a woman presenting with severe menorrhagia. Gynecol Oncol. 2003 Aug; 90(2): 348-52. doi: 10.1016/s0090-8258(03)00263-4. PMID: 12893198.
- Jameson CF. Angiomyoma of the uterus in a patient with tuberous sclerosis. Histopathology. 1990 Feb; 16(2): 202-3. doi: 10.1111/j.1365-2559.1990.tb01095.x. PMID: 2323745.
- Konichezky M, Reif R, Bukovsky I. Benign angiomyoma of the uterus with unusual macroscopic appearance. Int J Gynaecol Obstet. 1980 Jul-Aug; 18(1):4-6. doi: 10.1002/j.1879-3479.1980.tb00230.x. PMID: 6106598.
- Sahu L, Tempe A, Agrawal A. Angioleiomyoma of uterus. J Obstet Gynaecol. 2012 Oct; 32(7): 713-4. doi: 10.3109/01443615.2012.702148. PMID: 22943735.