

Case Report

Angioleiomyoma of Uterus Masquerading as Malignant Ovarian Tumor

Sweta Singh, Monalisha Naik, Narbadyswari Deep Bag¹, Susama Patra²

Departments of Obstetrics and Gynaecology,
¹Radio-diagnosis and
²Pathology, All India Institute of Medical Sciences, Bhubaneswar, Odisha, India

ABSTRACT

Uterine angioleiomyoma is rare. A 40-year-old nulliparous woman presented with heavy menstrual bleeding (HMB) for the past 2 years and mass per abdomen with severe dysmenorrhea for three cycles. She had received 8 units of packed cell transfusion outside. Clinical examination revealed a huge 32-week-sized abdominopelvic mass with irregular margins. Ultrasonography (USG) of the abdomen and pelvis showed a large solid cystic mass on the right side of the abdomen with a well-defined hypoechoic rounded lesion of size 5.7 cm × 5.0 cm, in the right lobe of the liver, with ovaries not being imaged separately. On color Doppler USG, there was moderate vascularity throughout. A provisional diagnosis of malignant ovarian tumor with hepatic metastasis was made. Her hemoglobin was 5.7 g/dl, and she had repeated episodes of HMB upon admission. She was transfused with 5 units of packed cells. Computed tomography (CT) showed a large fundal subserosal uterine fibroid on the right side, with a solid ovarian tumor measuring 5.0 cm × 4.5 cm on the left side, with ascitis, right-sided hydronephrosis, and a well-defined hypoechoic lesion in the right lobe of the liver, suggestive of hepatic hemangioma. Tumor markers were within normal limits. In view of discrepancy in clinical findings, ultrasound, and CT report, CT-guided biopsy of the huge mass was done which revealed leiomyoma, with no evidence of mitosis, pleomorphism, or malignancy. Laparotomy with total abdominal hysterectomy and bilateral salpingo-oophorectomy was done. Histopathology revealed an angioleiomyoma uterus. At 1-year follow-up, she was asymptomatic, and the liver mass was stable.

KEYWORDS: *Angioleiomyoma uterus, malignant ovarian tumor, vascular leiomyoma*

INTRODUCTION

Uterine angioleiomyoma is a rare variant of benign leiomyoma, which can mimic malignant ovarian tumor. Only a handful of cases have been reported worldwide. We report such a case which was successfully managed in our institute and discuss the challenges in diagnosis and management.

CASE REPORT

A 40-year-old nullipara presented with heavy menstrual bleeding (HMB) for the past 2 years and mass per abdomen with severe dysmenorrhea for the last three cycles. Her cycles lasted for 20–25 days, occurring at intervals of 30–60 days, associated with mass per abdomen and severe dysmenorrhea for 3 months. She

had received 8 units of packed cell transfusion outside for HMB with severe anemia. Ultrasonography (USG) done 1 month before showed a large, lobulated, heterogeneous space-occupying lesion in the abdomen, located posterior to urinary bladder, extending from pelvis to epigastric region, suggestive of malignant ovarian tumor. Her cycles 2 years back were regular with average flow and no dysmenorrhea.

On general examination, there was pallor. Breast and thyroid examinations were within normal limits.

Address for correspondence: Dr. Sweta Singh,

Department of Obstetrics and Gynaecology, All India Institute of Medical Sciences, Bhubaneswar - 751 019, Odisha, India.

E-mail: swetsingh@hotmail.com

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Singh S, Naik M, Bag ND, Patra S. Angioleiomyoma of uterus masquerading as malignant ovarian tumor. *J Mid-life Health* 2017;8:145-7.

Access this article online	
Quick Response Code: 	Website: www.jmidlifehealth.org
	DOI: 10.4103/jmh.JMH_47_17

Abdominal and bimanual examination revealed a huge abdominopelvic mass, corresponding to 32 weeks of gestation, which was firm, nontender, with restricted mobility and irregular margins, occupying all fornices. USG was repeated at our institute, which showed a large solid-cystic mass on the right side of abdomen [Figure 1a], with right-sided hydronephrosis, and a well-defined hypoechoic rounded lesion of size 5.7 cm × 5.0 cm in the right lobe of liver. On color Doppler USG, there was moderate vascularity throughout [Figure 1b]. A provisional diagnosis of malignant ovarian tumor with hepatic metastasis was made. Her hemoglobin was 5.7 g/dl, and she had repeated episodes of heavy bleeding per vaginum after admission. She was transfused with 5 units of packed cell transfusion preoperatively. Her serum thyroid-stimulating hormone was 13.10 U, anti-thyroid peroxidase antibody >1300 U/ml, and she was started on tablet levothyroxine 50 µg daily. In view of suspected malignancy, computed tomography (CT) was done which showed a large fundal subserosal uterine fibroid on the right side [Figure 1c], with a solid ovarian tumor measuring 5.0 cm × 4.5 cm on the left side, with ascitis, and a well-defined hypoechoic lesion in the right lobe of liver, suggestive of hemangioma [Figure 1d]. Tumor markers were within normal limits, with cancer antigen (CA)-125 being 19.3U/ml, CA-19-9 being 10.42 U/ml, alphafetoprotein 7.8 ng/ml, carcinoembryonic antigen 1.27 ng/ml, human chorionic gonadotropin 2.8 mIU/ml. In view of discrepancy in clinical findings, ultrasound and CT report, CT guided biopsy of the mass was done which revealed leiomyoma, with no evidence of malignancy.

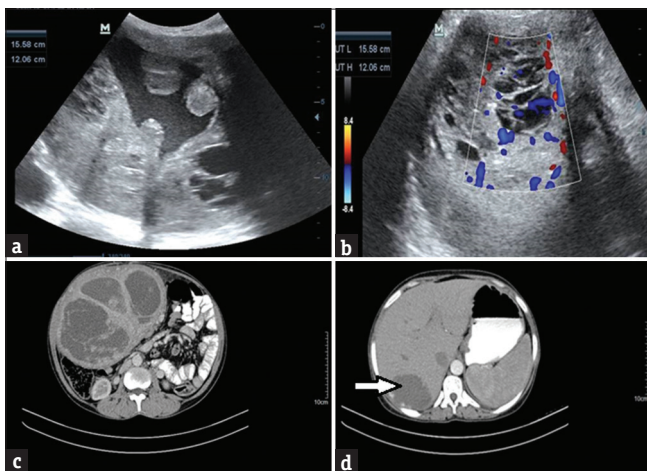


Figure 1: (a) Ultrasonography abdomen and pelvis showing a large solid-cystic mass on right side of abdomen, mimicking malignant ovarian tumor. (b) Color Doppler ultrasonography showing moderate vascularity throughout the mass (as opposed to the peripheral arrangement of vessels in fibroid uterus). (c) Computed tomography scan of abdomen showing large fundal subserosal fibroid on right side. (d) Computed tomography scan of abdomen showing a well-defined hypoechoic lesion (arrow) in the right lobe of liver posteriorly, suggestive of hemangioma

Laparotomy was performed under general anesthesia. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was done [Figure 2a] with moderate blood loss. The solid ovarian tumor seen on CT scan turned out to be fibroid of the left round ligament [Figure 2b]. Gross specimen weighed 5.1 kg. Patient was transfused with 3 units of packed cells peri-operatively. Postoperative period was uneventful.

Gross specimen measured 26 cm × 26.5 cm × 18 cm, with cut-section showing dilated uterine cavity containing blood clots. Multiple fibroids were present in intramural, subserous and submucous location. On microscopy, numerous interspersed vascular channels were noted [Figure 2c]. There no evidence of mitosis, pleomorphism or malignancy. Final diagnosis of leiomyomata uterus with large anterior fibroid showing features of angioleiomyoma was made. Patient is on follow-up since the last 1.5 years, and is asymptomatic with no recurrence and liver mass is stable.

DISCUSSION

Angioleiomyoma, also known as vascular leiomyoma, is a rare variant of benign leiomyoma, occurring in 0.34%–0.40% cases of uterine leiomyomas.^[1] Microscopically, three histological types are distinguished (1) capillary type with narrow vessels interlaced with thick fascicles of smooth muscles (2) venous type composed of thick vessels interspersed with fascicles of smooth muscles and (3) cavernous type with widened vessels and lesser amount of smooth muscles.^[1] Our case appeared to be cavernous type of uterine angioleiomyoma.

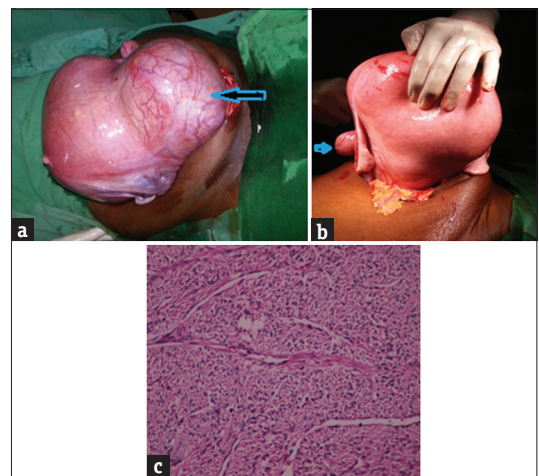


Figure 2: (a) Intra-operative picture, arrow pointing to large anterior subserous fibroid, which turned out to be vascular leiomyoma. (b) Intra-operative picture, arrow pointing to fibroid of left side round ligament, which was mistaken for solid ovarian tumor of left side on computed tomography; bilateral ovaries are seen to be normal. (c) Photomicrograph showing interlacing fascicles of spindle cells with numerous interspersed vascular channels

The exact aetiopathogenesis is not well known. Angioleiomyoma usually develops in the third to sixth decades. Till 2014, only 16 cases of uterine angioleiomyoma had been described in medical literature.^[2]

Most women with angioleiomyoma present with abnormal and sometimes massive bleeding per vaginum, along with severe dysmenorrhoea. HMB has been proposed to be due to dysregulation of growth factors like basic fibroblast growth factor and their receptors, which affects the venous plexuses in the leiomyoma and regulates the process of angiogenesis.^[3,4] Severe anemia may result, as seen in our case also, necessitating 8 units of packed cell transfusion and another 8 units elsewhere. Dysmenorrhea results probably due to local ischemia from vessel contraction.^[5] Women with angioleiomyoma have a greater risk of life-threatening complications such as spontaneous rupture due to high vascularity of these lesions.^[5] It has also been suggested that these tumors are derived from arteriovenous anastomosis.^[5] Consumptive coagulopathy secondary to large degenerated angioleiomyoma in a 38-year-old nulligravida has also been described.^[6] Angioleiomyoma may undergo degenerative changes, with large cavernous deformation of the vascular spaces.^[7]

Preoperative differentiation of angioleiomyoma with ovarian tumor is difficult due to frequent degenerative changes in the course of uterine angioleiomyoma.^[2] In all the cases reported thus far, a definitive diagnosis was made only after histopathologic examination of the operative specimen.^[2] Gross picture mimics conventional leiomyoma, except for the presence of blood filled spaces in angioleiomyoma. The ultrasonographic picture in most cases is indistinguishable from that of a conventional leiomyoma.^[2] Angioleiomyoma should be considered if CT scan reveals the presence of prominent tortuous vascular-like enhancing structures in a well-demarcated soft tissue mass arising from the uterus.^[3] However, a preoperative diagnosis is not always possible as

large-sized lesions may be predominantly solid with loss of fat plane with the uterine myometrium, and it may be difficult to visualize the ovaries in these cases, thereby mimicking ovarian neoplasms.^[8] In the majority of cases, surgical excision with hysterectomy is the treatment of choice.

CONCLUSION

Uterine angioleiomyoma is rare, with only a handful of cases reported in literature. It causes HMB and severe dysmenorrhea, with no accurate preoperative imaging. Surgical excision offers cure. This case is being reported for its rarity and challenges in diagnosis and management.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Weiss SW, Goldblum JR, editors. Benign tumors of smooth muscle. In: Enzinger and Weiss's Soft Tissue Tumors. 4th ed. St. Louis, MO: The CV Mosby Co.; 2001. p. 699-700.
2. Garg G, Mohanty SK. Uterine angioleiomyoma: A rare variant of uterine leiomyoma. Arch Pathol Lab Med 2014;138:1115-8.
3. Hsieh CH, Lui CC, Huang SC, Ou YC, ChangChien CC, Lan KC, *et al.* Multiple uterine angioleiomyomas in a woman presenting with severe menorrhagia. Gynecol Oncol 2003;90:348-52.
4. Stewart EA, Nowak RA. Leiomyoma-related bleeding: A classic hypothesis updated for the molecular era. Hum Reprod Update 1996;2:295-306.
5. Culhaci N, Ozkara E, Yüksel H, Ozsunar Y, Unal E. Spontaneously ruptured uterine angioleiomyoma. Pathol Oncol Res 2006;12:50-1.
6. Handler M, Rezaei F, Fless KG, Litinski M, Yodice PC. Uterine angioleiomyoma complicated by consumptive coagulopathy. Gynecol Oncol Case Rep 2012;2:89-91.
7. Agorastos T, Dinas K, Patsiaoura K. Cystic degenerated angioleiomyoma mimicking ovarian pathology. Acta Obstet Gynecol Scand 2001;80:863-5.
8. Thomas S, Radhakrishnan L, Abraham L, Matthai A. Uterine angioleiomyoma with atypia, raised CA-125 levels, and pseudo-Meigs syndrome: An alarming presentation. Case Rep Pathol 2012;2012:519473.