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Case Report

Giant angioleiomyoma of uterus- an unknown entity

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ABSTRACT

Angioleiomyoma of uterus is a very rare benign tumor, originating from mesenchymal tissue. It usually develops in the lower extremities of middle-aged women and rarely affects other body parts. Uterus is an extremely rare location for angioleiomyoma. We report a rare case of giant angioleiomyoma detected in a 42-year-old woman who is nulliparous and came with a very prominent abdomen.

1. Introduction

Angioleiomyoma of the uterus, also known as vascular leiomyoma, is a very rare benign tumor, originating from the mesenchymal tissue and composed of smooth muscle cells and thick-walled vessels. Angioleiomyoma occurs more frequently in the lower extremities and rarely affects other body regions.[1] It represents the 0.34%-0.40% cases of uterine leiomyomas.[2] Uterine angioleiomyoma presents as a lower abdominal mass accompanied by dysmenorrhea and hypermenorrhoea.[3] On imaging, the diagnosis is very difficult, and the final diagnosis is usually formulated on histopathological examination. To our knowledge, 41 cases of uterine angioleiomyoma have been reported so far.[4,5]

2. Case Presentation

A 42 year female unmarried, nulligravida presented to gynae OPD with complaints of bleeding per vaginum and heaviness in lower abdomen since 2 years. On per abdomen examination abdomen was uniformly distended and umbilicus was centrally inverted. A smooth regular mass around 36 weeks size of gravid uterus was palpable. The mass was well circumscribed, uniformly enlarged and lower margin could not be felt. Her lab investigations were within normal limits. USG abdomen was suggestive of large hyperechoic abdominal mass measuring 30x13 cm in size suggestive of Uterine fibroid. CT Abdomen was suggestive of large uterine leiomyoma. Patient underwent exploratory laparotomy and surgeon proceeded total abdominal hysterectomy with bilateral salpingectomy with right oophorectomy. Sample was sent for histopathological examination. Grossly received a uterus with cervix with right ovary and fallopian tube. Left fallopian tube was sent separately. Specimen of uterus with cervix measured 25x23x16 cm(Figure 1A). Externally the uterus was enlarged, distorted, oedematous with areas of haemorrhage. On cut section and serial slicing a large mass(leiomyoma/leiomyosarcoma) identified measuring 22x20x14 cm. Tumour shows variegated appearance with areas of haemorrhage(Figure 1B), nodular areas necrosis,and myxoid areas. Numerous blood vessels identified with clot in the lumen. Extensive sampling was done. Right fallopian tube measured 7 cm in length. Right ovary measured 5x3.5x2 cm. Cut section revealed cystic and haemorrhagic areas. Left fallopian tube measured 3.5 cm in length.

Microscopically, Endometrium showed late secretory phase. Myometrium showed a well circumscribed

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moderately cellular spindle cell tumour (Figure 1C) composed of interlacing fascicles of spindle to plump cells with minimal nuclear pleomorphism. There was presence of numerous thin dilated cavernous type blood vessels with focal area of infarct(Figure 1D). Blood vessels showed thrombi in variable stages. In addition edema, hyaline, cystic, myxoid degenerations and areas of haemorrhage were also noted(Figure 1E,F,G). No necrosis/mitosis was seen in the section studied for examination after extensive sampling. Cervix showed chronic non specific cervicitis with Nabothian follicles. Right fallopian tube was unremarkable with Walthard cell nests and right ovary showed corpus luteum haemorrhagicum with ovarian edema. Left fallopian tube showed salpingitis isthmica nodosa. Histopathological diagnosis was give as Giant angioleiomyoma.

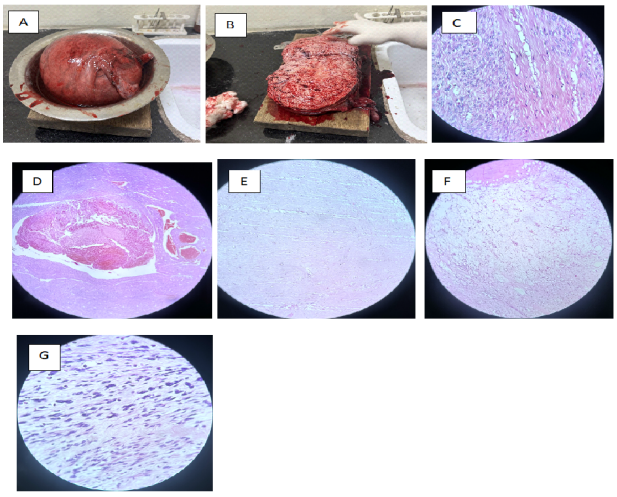

Figure 1: A) Gross Specimen of uterus with cervix measured 25x23x16 cm. Externally the uterus was enlarged, distorted, oedematous with areas of haemorrhage. **B)**Tumour shows variegated appearance with areas of haemorrhage. **C)**Myometrium shows a moderately cellular spindle cell tumour composed of interlacing fascicles of spindle to plump cells (10x). **D)**Numerous thin dilated cavernous type blood vessels and Blood vessels show thrombi in variable stages (10x). **E)**Hyaline degeneration(10x). **F)** Myxoid degeneration(10x). **G)** Degenerative atypia in the nucleus(40x).

3. Discussion

Uterine angioleiomyoma is an extremely rare and unique variant of leiomyom It usually occurs in middle-aged females. The age range is from 30 years to 69 years. Presenting symptoms in most of the cases are heavy menstrual bleeding, discomfort or pain in abdomen, distension of abdomen and/or abdominal mass.[6] It is a well-circumscribed mass arising from the uterus and largest dimension ranges from 4cm to 30 cm, but only few cases have been reported of size more than 20cm.[7] These can be found in the subserosa, submucosa or can be intramural.[8,9] The cut surfaces are usually white and whorled with or without congested or haemorrhagic areas, or they may exhibit a variegated appearance with pinkish-brown and grey areas. Based on the variable relationship among smooth muscles and vascular cavities, angioleiomyomas are classified into 3 histologic types: cavernous type, capillary or solid type and venous type.[10] In this case it was of cavernous type. Usually there are no features suggestive of mitosis, nuclear atypia, pleomorphism, or necrosis on microscopy. Angioleiomyoma may undergo degenerative changes, with large cavernous deformation of the vascular spaces.[1] Other changes that have been described in this tumor are myxoid change, edema, and hyalinization of the stroma, as well as fibrin deposition in the vessel wall.[11] The macroscopic features of AL overlap with those of a conventional LM, except for the presence of blood-filled spaces in AL. The tumor cells are immunoreactive for smooth muscle actin, desmin, h-caldesmon, and progesterone receptor, with a low Ki-67 labelling index. Because these lesions are vascular, they may undergo spontaneous rupture and pose a life-threatening emergency, especially in pregnancy. There are no specific imaging findings; therefore, a preoperative diagnosis is extremely difficult.[1] In most cases, the surgical treatment is hysterectomy with or without resection of appendages; myomectomy, when technically possible, is rare, and it is considered only to preserve fertility.[12]

4. Conclusion

Angiomyolipoma is a distinct variant of leiomyoma, with unusual morphology and immunohistochemistry findings. It is difficult to differentiate this from other smooth muscle neoplasm because it has no specific imaging findings and hence pre-operative diagnosis is less likely. Due to increasing incidence of this entity and clinical relevance, it should be included as a distinct variant of leiomyoma in the WHO classification.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Garg G, Mohanty SK. Uterine angioleiomyoma: a rare variant of uterine leiomyoma. *Arch Pathol Lab Med*. 2014;138(8):1115–8.
2. Singh S, Naik M, Bag ND, Patra S. Angioleiomyoma of uterus masquerading as malignant ovarian tumor. *J Midlife Health*. 2017;8(3):145–7.

3. Sato H, Murakami K, Fujishima R, Otani T, Sakai K, Nishio K, et al. Uterine angioleiomyoma with disseminated intravascular coagulation: a case report. *BMC Women's Health*. 2023;23(1):157.
4. Seth A, Mathur A. Angioleiomyoma of uterus and cervix: a rare report of two cases. *J Midlife Health*. 2021;12(2):179–84.
5. Gorginzadeh M, Tahermanesh K, Hanjani S, Khorshidizadeh A, Allahqoli L, Sadeghipour A, et al. Uterine intracavitary angioleiomyoma. *J Obstet Gynaecol*. 2020;40(8):1186–8.
6. 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(2):348–52.
7. Manchanda R, Dave A, Goyal K, Goyal R. Giant Angioleiomyoma of Uterus: A Rare Case with its Management. *J MAR Gynecol*. 2022;3(4).
8. Hakverdi S, lu KD, Güngören A, Yaldiz M, Hakverdii AU. Multiple uterine angioleiomyomas mimicking an ovarian neoplasm: A case report. *EurJ Gynaec Oncol*. 2009;30:592–4.
9. Konichezky M, Reif R, Bukovsky I. Benign angiomyoma of the uterus with unusual macroscopic appearance. *Int J Gynaecol Obstet*. 1980;18(1):4–6.
10. 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;27(3):69–71.
11. Mccluggage WG, Boyde A. Uterine angioleiomyomas: a report of 3 cases of a distinctive benign leiomyoma variant. *Int J Surg Pathol*. 2007;15(3):262–5.
12. Pierro A, Rotondi F, Cilla S, Ninno MD, Mattoni M, Berardi S, et al. Giant angioleiomyoma of uterus: a case report with focus on CT imaging. *Radiol Case Rep*. 2018;13(2):371–5.

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