International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Yadav M et al. Int J Reprod Contracept Obstet Gynecol. 2025 Dec;14(12):4380-4382
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/ 10.18203/2320-1770.ijrcog20253914

Case Report

Angioleiomyoma of uterine cervix – a common tumour at rare site

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Received: 02 March 2025
Revised: 01 May 2025
Accepted: 25 October 2025

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ABSTRACT

Angioleiomyoma of the uterine cervix is a rare benign tumor of this site characterized by smooth muscle proliferation intermixed with prominent vascular structures. At cutaneous locations it presents as painful nodule however cervical angioleiomyoma clinically manifests with non-specific, mimicking other gynecological conditions such as fibroids or adenomyosis. No specific clinical or radiological features are noted hence histopathology remains the gold standard for diagnosis and sometimes immunohistochemistry is helpful in confirming smooth muscle origin. This case report details a case of uterine cervical angioleiomyoma in a 45-year-old woman presenting with abnormal excess uterine bleeding and pelvic pain. On radiology a diagnosis of cervical fibroid polyp was rendered. Following which surgical excision of the mass revealed the diagnosis, confirmed by histopathological and IHC evaluation. While angioleiomyoma is rare at this site may mimic benign or vascular tumors clinically or even microscopically. Hence correct and timely diagnosis is essential for appropriate clinical management.

Keywords: Angioleiomyoma, Tumour, Gynecological conditions

INTRODUCTION

Angioleiomyoma is a benign neoplasm originating from smooth muscle cells with a prominent vascular component. emerges as a medical rarity, primarily manifesting in adults between their 40s and 60s. While commonly seen in the lower extremities, head, neck, and trunk, its presence in the female lower genital tract, especially the uterus, is a medical curiosity. Among these, cervical angioleiomyoma shines as an exceptionally rare gem, with a mere handful of cases known to the medical world.[1]

Frequently occurring in the extremities and head-and-neck area, angioleiomyoma stands out for its rarity in the uterus, cervix, ovary, and broad ligament. This tumor, believed to be a morphological offshoot of the more common uterine leiomyoma, has only a few reported instances, making each case a valuable addition to medical literature.[2]

Known alternatively as vascular leiomyoma, this tumor is a unique variant within the realm of uterine smooth muscle neoplasms. It is intricately composed of smooth muscle cells intertwined with vascular components. The real challenge in identifying angioleiomyoma lies in its elusive nature; it often masquerades as a typical leiomyoma in radiological evaluations and gross examinations, revealing its true identity only under the discerning eye of microscopic examination.[3] The vascular nature of this tumor adds a layer of complexity, as it is prone to spontaneous rupture, potentially escalating into a life-threatening situation. Each diagnosis of angioleiomyoma is not just a medical victory in pinpointing this elusive tumor, but also a crucial step in averting potential emergencies.[3] The study and understanding of angioleiomyoma not only enrich our knowledge of rare tumors but also highlight the intricacies and surprises within human pathology. In this report, we present a case of angioleiomyoma of the uterine cervix because of its extreme rarity.

CASE REPORT

A 45-year-old female was presented to our gynecological outpatient department (OPD) with a recurrent urinary retention and hypermenorrhea. Her vitals were stable and she was P3L3 with all normal vaginal deliveries. On vaginal examination large protruding mass was seen on the right sided wall of the uterine cervix, and a further physical examination revealed that it was within normal limits. For further evaluation of the cervical mass, blood tests including tumor markers and radiological investigations, were performed. The patient's blood indices were as follows: hemoglobin 8.3 g/dl, LDH (175 u/l) and other indices including AFP, CEA, beta HCG, carbohydrate antigen 19-9 (CA19-9) and CA125 were in the normal range.

A whole abdomen ultrasonography (US) was performed and a well-demarcated, heterogeneous, large echogenic lesion measuring 11 cm in endocervical canal and vagina with a vascular pedicle likely fibroid polyp. The magnetic resonance imaging (MRI) pelvis, showed that the mass was heterogeneously signal intensity measuring 10.3×9×4.3 cm and located in uterine cervix and lower uterine region of uterus with exogenous component likely prolapsed cervical fibroid polyp. A total abdominal hysterectomy with bilateral salpingectomy were performed. At histopathological examination, macroscopically, a globular cervical mass with ectocervix all together measuring 11.8×9.5×4.6 cm. outer surface of mass is congested and cut surface show homogenous gray white areas with foci of hemorrhage at places (Figure 1a). There was no evidence of necrosis in the specimen. Uterus measures 6.8×62×40 cm and grossly unremarkable with bilateral tubes measuring 19×8 and 18×7 cm respectively (Figure 1a).

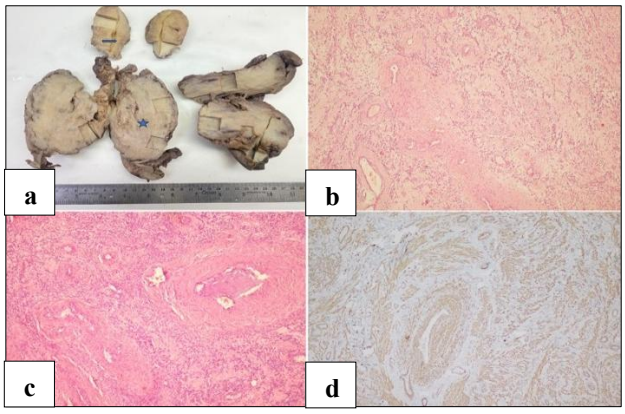

Figure 1: (a) Gross of total abdominal hysterectomy specimen with grey coloured circumscribed growth at cervix (asterisk), (b) highly vascular benign spindle cell tumor without areas of necrosis, H and E, 100x, (c) spindle cells are forming fascicle's and seem to originate from the blood vessel wall, H and E, 100x, and (d) cytoplasmic positivity for H-caldesmon, immunohistochemistry, 400x.

Microscopically, the tumor was well circumscribed and composed by spindle cells arranged in vague fascicles and numerable proliferating thick-walled and thin wall blood vessels (Figure 1b and c). Areas of hyalinization and haemorrhage were also noted. No atypical cells or mitosis or necrosis seen. In immunohistochemical study, H-caldesmon was performed and tumor cells expressed diffuse cytoplasmic positivity (Figure 1d). Based on the results, the tumor was diagnosed as an angioleiomyoma of the cervix.

DISCUSSION

Angioleiomyoma is a rare subset of leiomyomas, comprising smooth muscle cells interspersed with thick-walled blood vessels. While it is more commonly seen in subcutaneous tissues of the extremities, its occurrence in the uterine cervix is exceedingly uncommon.[2] This rarity poses challenges in preoperative diagnosis and management, as the clinical presentation is often non-specific, overlapping with more common conditions like uterine fibroids, polyps, or adenomyosis.[4] Symptoms of cervical angioleiomyoma can include abnormal uterine bleeding, pelvic pain, or a palpable cervical mass, although some patients may be asymptomatic.[5]

Imaging modalities such as ultrasound and magnetic resonance imaging (MRI) are instrumental in delineating the lesion. On ultrasound, these tumors are usually hypoechoic and well-circumscribed, while MRI reveals a hypervascular mass with smooth borders, aiding in differentiating it from infiltrative or malignant lesions. However, the vascularity seen in imaging can mimic other conditions, such as hemangiomas or cervical malignancies, necessitating histological confirmation.[7-9] Histologically, angioleiomyomas are characterized by well-demarcated proliferation of smooth muscle cells with intervening thick-walled blood vessels. These tumors are benign, but the vascular component can sometimes be confused with angiosarcomas or hemangiopericytomas, emphasizing the importance of immunohistochemical analysis. Positive staining for smooth muscle actin (SMA) and desmin confirms smooth muscle origin, while negative staining for CD34 and other endothelial markers helps exclude pure vascular tumors.[4,6] The differential diagnosis for cervical angioleiomyoma includes leiomyoma a common benign smooth muscle tumors lacking prominent vascular structures, hemangiomas, vascular tumors composed of endothelial-lined spaces, usually positive for CD31 and CD34 and adenomyosis characterized by ectopic endometrial glands within the myometrium, often with less defined margins. Surgical excision is the definitive treatment for cervical angioleiomyoma, both for symptom relief and diagnostic confirmation.[3,6] Complete excision typically results in an excellent prognosis, as recurrence or malignant transformation is exceedingly rare. Follow-up is generally straightforward, focusing on symptom recurrence or imaging findings suggestive of residual or recurrent tumor.[1] The rarity of this condition means that most gynaecologists and pathologists may not

encounter it in routine practice. Increased awareness and detailed reporting of such cases are critical to expanding our understanding of the clinical and pathological spectrum of cervical angioleiomyomas.[2] Moreover, this case emphasizes the role of multidisciplinary collaboration, particularly between gynaecologists, radiologists, and pathologists, to ensure accurate diagnosis and optimal patient outcomes.

CONCLUSION

In conclusion, while angioleiomyomas of the uterine cervix are rare, their recognition is important for appropriate management. Histopathological evaluation remains indispensable for definitive diagnosis, and surgical excision provides curative treatment. Further studies and case reports can help refine diagnostic approaches and management strategies for this uncommon entity.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Yadav M, Singh M, Tyagi S, Rajput R. Angioleiomyoma of uterine cervix – a common tumour at rare site. Int J Reprod Contracept Obstet Gynecol 2025;14:4380-2.