International Journal of Women's Health

Dovepress
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CASE SERIES

Torsion of Angioleiomyoma in the Female Genital Tract: A Rare Report of Two Cases

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Background: Angioleiomyoma (ALM) is a rare, benign tumor involving the peripheral soft tissues. Preoperative diagnosis of ALM is challenging, since there are no characteristic clinical or imaging findings. The objective of this article is to present two rare cases of ALM with torsion involving the female genital tract and review published similar cases.

Case Presentation: We report two cases of ALM with torsion originating from the uterus and mesosalpinx, respectively. The two cases presented with abdominal dull pain or a palpable abdominal mass, and imaging findings revealed a large cystic mass, which was suspected to be an ovarian neoplasm. During the surgical procedure, it was found that the mass had undergone torsion. The final histological and immunohistochemical reports of the resected masses validate the diagnosis of ALM.

Conclusion: To our knowledge, this is the first report of torsion of ALM originating from the female genital tract. ALM poses a significant diagnostic challenge in cases of a pelvic cystic mass with torsion, as it is frequently misdiagnosed as an ovarian tumor. Surgical excision of the mass while preserving the uterus and adnexa should be strongly recommended for young patients, as ALM has an excellent prognosis.

Keywords: angioleiomyoma, cystic mass, torsion, surgical excision

Introduction

Angioleiomyoma (ALM) primarily occurs in the subcutis or dermis of the extremities, formed by smooth muscle cells and thick-walled vascular vessels.[1] To date, the exact etiology of angioleiomyoma remains unclear. According to the 2020 World Health Organization Classification of Soft Tissue Tumors, angioleiomyoma belongs to the pericytic (perivascular) tumor group. It is supposedly a distinct variant of leiomyoma which is the most common uterine neoplasm. In contrast to the usual leiomyoma, angioleiomyoma is also composed of bland smooth muscle cells but is distinguished by a prominent vascular component comprised of numerous, evenly distributed blood vessels. It is hypothesized that while usual leiomyomas originate from myometrial smooth muscle cells, angioleiomyomas derive from perivascular smooth muscle cells. However, ALM involving the female genital tract, including uterine corpus, cervix, broad ligament and ovary, is rare and only case reports and occasional studies have been previously documented in the literature.[2–7] Pelvic localization is extremely rare with only about 100 cases described in the literature. Torsion of a pedunculated subserosal leiomyoma is uncommon; nevertheless, a considerable number of cases have been documented in the literature. And torsion of pedunculated leiomyoma is considered an acute surgical emergency due to the high risk of ischemic gangrene and peritonitis. However, there has never been a report regarding the torsion of ALM originating from the female genital tract. Due to the lack of specific clinical and imaging manifestations preoperatively, ALMs with torsion are often misdiagnosed as ovarian tumors, a diagnostic challenge compounded by the fact that the most common cause of pelvic torsion is an ovarian mass. Herein, we first describe two cases of ALM with torsion originating from the female genital tract (the uterus and mesosalpinx, respectively), presenting as a huge pelvic cyst mimicking ovarian tumors.

Received: 22 August 2025
Accepted: 5 November 2025
Published: 21 November 2025

Case Presentation

Case 1

A 35-year-old female, weighing 64kg and 166cm in height, was admitted to our hospital due to the sudden onset of diffuse abdominal dull pain for five days. Physical examination revealed local abdominal tenderness without rebound tenderness. Laboratory tests showed slightly elevated C-reactive protein level (14.3mg/L, normal value < 5mg/L) with normal white blood cell counts. Transabdominal sonography revealed a cystic mass (8.6 x 7.6×6.8 cm) posterior to the uterus, closely adjacent to the left ovary (Figure 1A). The cyst showed hypoechoic content with short linear strong echoes. No obvious blood flow signal was observed inside or around the cyst. The pelvic magnetic resonance imaging (MRI) showed a cystic mass with a thin, smooth wall. T2-weighted sequence showed diffuse hyperintensities of the mass with focal hypointensities adjacent to the uterus suggesting a pedicle with torsion in the coronal (Figure 1B) plus axial (Figure 1C) planes. DWI sequence showed heterogeneous hypointensities (Figure 1D) and contrast-enhanced T1-weighted sequence showed an enhancement of the cyst wall (Figure 1E). The tumor marker levels were all within normal range limits.

Ovarian cyst with torsion was suspected preoperatively and emergent surgery was performed. Laparoscopic exploration showed a large, well-circumscribed, black purplish cyst with complete torsion (540 degrees), originating from the posterior wall of the uterus (Figure 1F). Excision of the mass was done smoothly. The content of the cyst consisted of about 500cc of bloody fluid.

Definitive histologic examination revealed a small number of smooth muscle cells and abundant thick-walled vessels with diffuse hemorrhage and necrosis. Immunohistochemically, this case was diffusely positive for smooth muscle antigen (SMA) in smooth muscle cells. CD31 and CD34 were positive in the endothelial cells (Figure 2). These findings validate the diagnosis of ALM. The patient remains disease-free after six months of follow-up.

Case 2

An 18-year-old virgin, weighing 46kg and 151cm in height, was admitted to our hospital for evaluation of a palpable abdominal mass of 3 months' duration. She did not have any other complaints, such as dysmenorrhea, menorrhagia, abdominal pain or change in bowel habits. Physical examination revealed a large pelvic mass with a smooth surface, and no tenderness was identified. Transabdominal ultrasound showed a giant cystic mass (14.5 x 13.8×9.6 cm) anterior to the

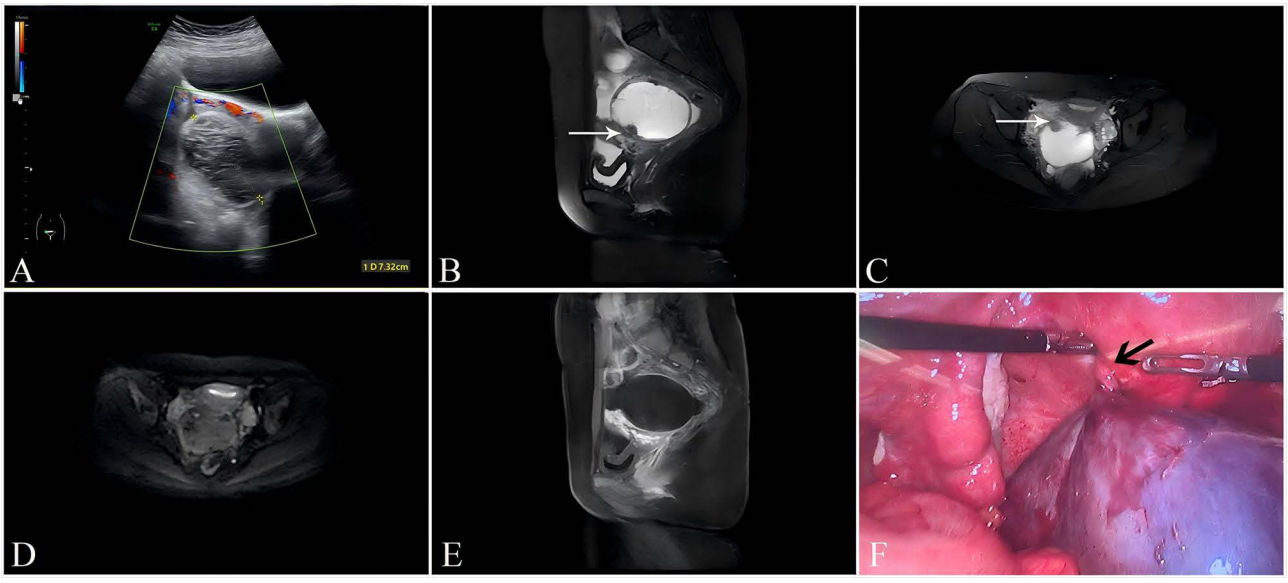

Figure 1 (**A**)Transabdominal sonography revealed a cystic mass (8.6 x 7.6×6.8 cm) posterior to the uterus, closely adjacent to the left ovary, and a hypoechoic cystic content with short linear strong echoes; (**B**) T2-weighted sequence showed diffuse hyperintensities of the mass with focal hypointensities adjacent to the uterus suggesting a pedicle with torsion (*white arrow*) in the coronal plane; (**C**) T2-weighted sequence showed diffuse hyperintensities of the mass with focal hypointensities adjacent to the uterus suggesting a pedicle with torsion (*white arrow*) in the axial plane; (**D**) DWI sequence showed heterogeneous hypointensities; (**E**) Contrast-enhanced T1-weighted sequence showed an enhancement of the cyst wall. (**F**) Laparoscopic exploration showed 540 degrees torsion (*black arrow*) of a cyst originating from the posterior wall of uterus.

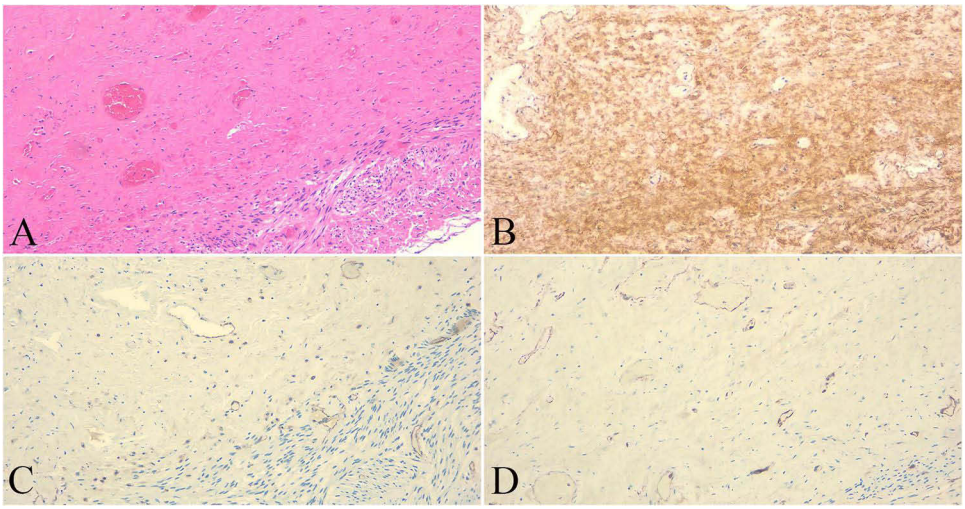

Figure 2 (**A**) Histological features showed a small number of smooth muscle cells and abundant thick-walled vessels with diffuse hemorrhage and necrosis (HE×100); (**B**) Immunohistochemical features showed SMA was positive in smooth muscle cells (×100); (**C**) CD31 was positive in the endothelial cells of the vessel walls (×100); (**D**) CD34 was positive in the endothelial cells of the vessel walls (×100).

uterus (Figure 3A). The cyst showed hypoechoic content with short linear strong echoes, which were divided into a "grid-like" structure. No obvious blood flow signal was observed inside or around the cyst. A computerized tomography (CT) was suggested, but declined by the girl's parents. On magnetic resonance imaging (MRI), it was observed as a cystic mass with a thin, smooth wall T2-weighted sequence showed slight hyperintensities with marked spot-like or strip-like hypointensities (a dark reticular sign) (Figure 3B), DWI sequence showed hypointensities and "grid-like" structure (Figure 3C), and the contrast-enhanced T1-weighted sequence showed septa within the mass with vascular enhancement (Figure 3D). No signs of ascites or pelvic dissemination were observed. The tumor marker levels

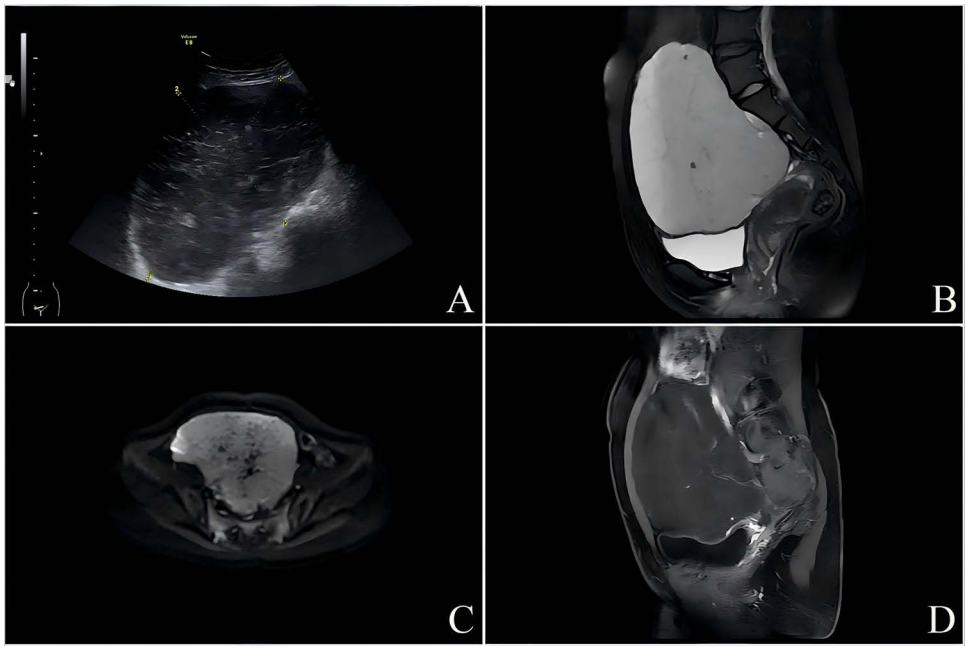

Figure 3 (**A**) Transabdominal ultrasound showed a giant cystic mass (14.5 x 13.8×9.6 cm) anterior to the uterus, and a hypoechoic cystic content with short linear strong echoes ("grid-like" structure); (**B**) T2-weighted sequence showed slight hyperintensities with marked spot-like or strip-like hypointensities (a dark reticular sign); (**C**) DWI sequence showed hypointensities and "grid-like" structure; (**D**) Contrast-enhanced T1-weighted sequence showed septa within the mass with vascular enhancement.

https://doi.org/10.2147/IJWH.S562540

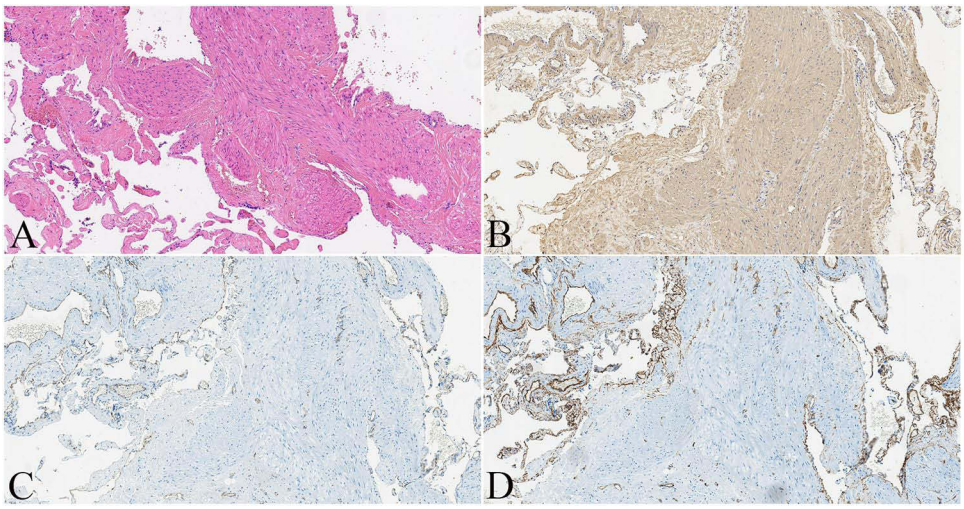

Figure 4 (**A**) Histological features showed a small number of smooth muscle cells and abundant vessels with thick hyalinized walls and several dilated lumina (HE×100); (**B**) Immunohistochemical features showed SMA was positive in smooth muscle cells (×100); (**C**) CD31 was positive in the endothelial cells of the vessel walls (×100); (**D**) CD34 was positive in the endothelial cells of the vessel walls (×100).

were all within normal limits (CA-125 21.0 U/mL, CA-199 20.0 U/mL, CA153 8.3 U/mL, CEA 0.7 ng/mL, and AFP 2.5ng/mL). A chest radiograph was normal. All blood routine and blood biochemistry data were within the normal range.

A clinical diagnosis of ovarian tumor was made preoperatively and scheduled surgery was performed. At the time of exploratory laparotomy, there was no ascites, but a giant, pink, well-circumscribed, cystic mass with incomplete torsion (180 degrees), originating from the right mesosalpinx. The cystic mass was resected, preserving the right ovary and fallopian tube. The content of the cyst consisted of about 1500cc of bloody fluid. The patient experienced a straightforward recovery following the operation. Regrettably, the tumor was assumed to be an ordinary Müllerian duct cyst or mucinous cystadenoma before resection; thus, pictures were not recorded during the surgery.

Definitive histologic examination of the mass showed smooth muscle and numerous thick-walled vessels with several dilated lumina. No significant pleomorphism, mitotic figures, or coagulative necrosis were observed. Immunohistochemically, this case was diffusely positive for SMA in fascicles encircling blood vessels, as well as in the smooth muscle component in the background. CD31 and CD34 were positive in the endothelial cells (Figure 4). These findings validate the diagnosis of ALM. The patient remains disease-free after seven years of follow-up.

Discussion

ALM, arising from the vascular smooth muscle, is a rare neoplasm that is considered a distinct variant of leiomyoma. Although leiomyoma is the most common uterine neoplasm, ALM involving the female genital tract is not frequently encountered and predominantly occurs in the subcutis or dermis of the extremities. To date, only approximately 100 cases involving the female genital tract have been reported in the English literature. Of these, the uterus is the most frequently involved site.[2,8,9]

Uterine ALM has been described more frequently in women ranging from 30–69 years of age and cases usually present with abnormal uterine bleeding, or pelvic mass.[2] Some patients experienced abdominal distension and abdominal pain. The clinical manifestations of ALM are non-specific and analogous to those of common leiomyoma, which are closely related to the location of the lesions. And there have been case reports of uterine ALM with spontaneous rupture[10] or disseminated intravascular coagulation.[11] Torsion is a rare acute complication of pedunculated subserous uterine leiomyoma, causing acute abdomen, but cases of torsion have not been reported in ALM. The risk factors contributing to torsion include the size of the mass and the characteristics of its stalk, which must be thin and long for rotation and torsion to occur. Our two cases of ALM with torsion are the first reported worldwide. The two cases both presented as a large mass with a thin and long pedicle, originating from the uterus and mesosalpinx, respectively. These morphological characteristics of the masses in our cases increase their propensity for torsion. Although a very few cases

of ALM in the ovary have been reported in the literature,[7,12,13] there has never been a report of an ALM with torsion originating from the mesosalpinx. In case 2, the 18-year-old patient presented with a palpable abdominal mass mimicking an ovarian neoplasm and exploratory laparotomy revealed a twisting of pedicle originating from the mesosalpinx.

Imaging techniques, such as ultrasound, CT, and MRI, have been performed in the perioperative study of patients with ALM. However, ALM has no specific imaging findings and is extremely difficult to distinguish from other masses, especially when it occurs in the female genital tract masquerading as an ovarian tumor or conventional leiomyoma. Despite this, a dark reticular sign on T2-weighted images appears to be a characteristic feature of angioleiomyoma in the extremities.[14] As in case 2, the patient presented as a cystic mass which was hyperintense on T2-weighted imaging (T2WI), with markedly spot-like or strip-like hypointensities inside the cyst, which may represent a specific imaging finding. This observation aligns with the dark reticular sign, suggesting a preoperative diagnosis of ALM. However, the MRI appearance can vary depending on the presence or absence of hemorrhage or infarction in torsion of ALM. In case 1, the complete torsion may contribute to the absence of this characteristic finding. In the study of 562 cases of ALM by Hachisuga, the most common type was the solid variant.[1] As in our cases, the blood-filled cystic masses were quite different from the cases reported in the literature which were usually described as a solid mass or leiomyoma-like mass rather than a cyst.

The gold standard for the diagnosis of ALM requires histological and immunohistochemical results. Histologically, ALM is composed of interlacing fascicles of monotonous spindled smooth muscle cells spinning around the abundant thick-walled blood vessels. Typically, there is no significant pleomorphism, mitotic figures, or coagulative necrosis present. Although case 1 suggested diffuse necrosis, this may be attributed to the complete torsion. Immunohistochemically, ALM shows positivity for smooth muscle antigen (SMA), desmin and h-caldesmon in smooth muscle cells. CD31 and CD34, on the other hand, highlighted the endothelial cells in the vessel walls. These special stains help differentiate ALM from other spindle cell neoplasms, such as fibroma, angiofibroma, angiomyolipoma and angiomyofibroblastoma.

Studies have shown that hysterectomy and complete resection of ALM are effective treatments of choice, depending on the patient's age, symptoms, location of the ALM and desire to preserve fertility. Although hysterectomy with or without bilateral salpingo-oophorectomy is the treatment of choice and was performed in most cases of uterine ALM, the role of tumor resection with preservation of the uterus or adnexa is essential since ALM is a benign tumor with an excellent prognosis. While ALM is a benign tumor associated with a good prognosis and a negligible risk of malignancy, long-term follow-up remains necessary because recurrence, though uncommon, can occur. We recommend that follow-up be scheduled every three months for the first year postoperatively, every six months for the second year, and annually thereafter.

Although our cases present singular characteristics, this study has several limitations that should be acknowledged. The primary constraints are the small number of cases and its reliance on case reports, which limit the generalizability of our findings and preclude drawing definitive conclusions. Therefore, future research involving larger, multi-center case series or comparative studies is necessary to validate and expand upon our observations.

Conclusion

ALM is a rare benign tumor. Preoperative diagnosis of ALM is extremely difficult, since there are no characteristic clinical or imaging findings. Postoperative pathology with immunohistochemical staining will confirm the diagnosis. It is important for the gynecologist and pathologist to recognize this uncommon benign tumor when it involves the female genital tract and surgical excision of the tumor, preserving the uterus and adnexa, should be strongly recommended for young patients, as ALM has an excellent prognosis. It is also important to recognize the need for emergent surgery in any patient presenting with acute abdominal pain and the clinical suspicion of torsion. The two cases highlight the complexity of the diagnosis of ALM with torsion.

Data Sharing Statement

The datasets used during the current study are available from the corresponding author on reasonable request.

Ethics Approval and Informed Consent

The study and publication of the two cases details were approved by the independent Clinical Research Ethics Committee of Women's Hospital, School of Medicine, Zhejiang University (IRB-20250034-R).

Consent for Publication

Written informed consents were obtained from the two patients for publication of this report.

Acknowledgments

We thank the two patients for allowing us to publish this report.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Disclosure

The authors declare that they have no competing interests.

References

1. Hachisuga T, Hashimoto H, Enjoji M. Angioleiomyoma. A clinicopathologic reappraisal of 562 cases. *Cancer*. 1984;54(1):126–130. doi:10.1002/1097-0142(19840701)54:1<126::aid-cncr2820540125>3.0.co;2-f
2. He S, Jiang J. Clinical characteristics and treatment outcomes of angioleiomyoma of the female genital tract: a retrospective cohort study. *BMC Womens Health*. 2024;24(1):479. doi:10.1186/s12905-024-03322-6
3. Seth A, Mathur A. Angioleiomyoma of uterus and cervix: a rare report of two cases. *J Midlife Health*. 2021;12(2):179–184. doi:10.4103/jmh.JMH_102_19
4. Lee J, Shin S, Kim JH, et al. Uterine cervical angioleiomyoma mimicking squamous cell carcinoma. *Diagnostics*. 2023;13(14):2370. doi:10.3390/diagnostics13142370
5. Kim H, Lee JJ, Choi Y, et al. Successfully removed uterine angioleiomyoma by robot-assisted laparoscopic myomectomy. *Obstet Gynecol Sci*. 2018;61(3):425–429. doi:10.5468/ogs.2018.61.3.425
6. Hwang YC, Park SY. A rare angioleiomyoma of the uterine cervix: a case report with peculiar MRI findings. *Taehan Yongsang Uihakhoe Chi*. 2022;83(3):693–698. doi:10.3348/jksr.2021.0127
7. Bouraoui S, El Amine El Hadj O, Rekik W, et al. First case of angioleiomyoma originating from the ovary of an adult woman. *Gynecol Obstet Invest*. 2010;70(1):8–10. doi:10.1159/000279310
8. Gupta M, Suryawanshi M, Kumar R, Peedicayil A. Angioleiomyoma of uterus: a clinicopathologic study of 6 cases. *Int J Surg Pathol*. 2018;26(1):18–23. doi:10.1177/1066896917731516
9. Sanchez-Iglesias JL, Capote S, Cubo-Abert M, et al. A giant superinfected uterine angioleiomyoma with distant septic metastases: an extremely rare presentation of a benign process and a systematic review of the literature. *Arch Gynecol Obstet*. 2019;300(4):841–847. doi:10.1007/s00404-019-05267-w
10. Culhaci N, Ozkara E, Yuksel H, Ozsunar Y, Unal E. Spontaneously ruptured uterine angioleiomyoma. *Pathol Oncol Res*. 2006;12(1):50–51. doi:10.1007/BF02893432
11. Sato H, Murakami K, Fujishima R, et al. Uterine angioleiomyoma with disseminated intravascular coagulation: a case report. *BMC Womens Health*. 2023;23(1):157. doi:10.1186/s12905-023-02292-5
12. Hsu TL, Changchien CC, Huang CC, Lin H. Angioleiomyoma originating from the ovary of an eleven-year-old premenarchal girl. *Gynecol Obstet Invest*. 2008;65(4):262–265. doi:10.1159/000113083
13. Lee SJ, Choi YS, Park KK. Ovarian angioleiomyoma: a case report. *Int J Clin Exp Pathol*. 2014;7(11):8235–8239.
14. Edo H, Matsunaga A, Matsukuma S, et al. Angioleiomyoma of the extremities: correlation of magnetic resonance imaging with histopathological findings in 25 cases. *Skeletal Radiol*. 2022;51(4):837–848. doi:10.1007/s00256-021-03888-4