

## Angiolipoleiomyoma of the Uterus: A rare case report and review of the literature

### *Uterin anjiolipoleiomyoma: Nadir bir olgu sunumu ve literatür taraması*

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### ABSTRACT

The aim of this study is to present a case of angiolipoleiomyoma (ALLM), a rare benign tumor of the uterus, compare it with similar cases in the literature, and contribute to the diagnosis, treatment, and classification process of this tumor.

A 40-year-old diabetic woman, gravida 4 para 4, presented to the emergency department with prolonged vaginal bleeding accompanied by dull lower abdominal pain. On admission, her hemoglobin level was 6.8 g/dL and hematocrit were 23.9%. Gynecological examination revealed a relaxed abdomen and a semi-mobile, non-tender fullness in the right adnexal region on bimanual palpation.

Further history indicated that the patient had been experiencing prolonged abnormal uterine bleeding as the primary symptom, with the dull pain emerging more recently. Following urgent blood transfusion, pelvic ultrasonography revealed a heterogeneous, hyperechoic cystic-solid mass located on the right side of the uterus.

Pelvic magnetic resonance imaging (MRI) showed a lobulated mass measuring 90×52 mm, slightly hyperintense on T1-weighted sequences, and demonstrating heterogeneous enhancement after contrast administration.

The mass was surgically removed via a Pfannenstiel incision. Histopathological examination confirmed the diagnosis of ALLM.

A comprehensive literature search was conducted using the keyword "angiolipoleiomyoma" in PubMed and Google Scholar up to June 2024. Animal studies were excluded from the review. As a result, 31 cases were identified, and diagnoses and classifications were evaluated according to the most recent histopathological criteria.

ALLM is a rare, typically asymptomatic, and benign tumor of the uterus. Definitive diagnosis is made through histopathological examination. The information from the literature and the presented case highlights the diagnostic characteristics of this tumor and contributes to future classification studies.

Note: This study was presented as a poster at the 15th Turkish-German Gynecology Congress, held on April 23–27, 2025, at Rixos Sungate, Antalya, Türkiye.

**Keywords:** Angiolipoleiomyoma, rare uterine tumor, pecoma

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## ÖZ

*Bu çalışmanın amacı, uterusun nadir görülen benign tümörlerinden biri olan anjiyolipoleiomyoma (ALLM) olgusunu sunmak, literatürdeki benzer vakalarla karşılaştırmak ve bu tümörün tanı, tedavi ve sınıflandırma sürecine katkı sağlamaktır.*

*40 yaşında, G4P4, diyabetik bir kadın hasta, uzun süredir devam eden vajinal kanama ve buna eşlik eden künt karakterde alt abdominal ağrı şikayetleri ile acil servise başvurdu. Başvuru anında yapılan tam kan sayımında hemogloblin düzeyi 6.8 g/dL, hematokrit %23,9 olarak saptandı. Jinekolojik muayenede batın rahat olup, sağ adneksiyel bölgede tuşeyle ağrısız, semimobil dolgunluk mevcuttu.*

*Anamnezde hastanın öncelikli şikayetinin uzun süredir devam eden vaginal kanama olduğu, buna son dönemde eklenen künt tarzda bir ağrının eşlik ettiği öğrenildi. Acil kan transfüzyonunun ardından yapılan pelvik ultrasonografide, uterusun sağ tarafına yerleşimli, hiperekojen, heterojen içerikli kist-solid nitelikte bir kitle tespit edildi.*

*İleri görüntüleme amacıyla yapılan pelvik manyetik rezonans (MR) incelemesinde, lobüle konturlu, T1A sekansında hafif hiperintens sinyal özellikleri gösteren, kontrast sonrası heterojen boyanma izlenen, yaklaşık 90×52 mm boyutlarında bir kitle saptandı.*

*Cerrahi olarak Pfannenstiel insizyonu ile çıkarılan kitle, histopatolojik olarak ALLM ile uyumlu bulundu.*

*Haziran 2024'e kadar "angioliipoleiomyoma" anahtar kelimesi kullanılarak PubMed ve Google Scholar veri tabanlarında kapsamlı bir literatür taraması yapılmıştır. Hayvan çalışmaları dışlanmıştır. Tarama sonucunda 31 olgu belirlenmiş, tanı ve sınıflandırmalar güncel histopatolojik kriterlere göre değerlendirilmiştir.*

*ALLM, uterusun nadir görülen, genellikle asemptomatik ve benign seyirli bir tümördür. Kesin tanı histopatolojik inceleme ile konur. Literatür bilgisi ve sunulan olgu, bu tümörün tanısal özelliklerine dikkat çekmekte ve ileride yapılacak sınıflandırma çalışmalarına katkı sağlamaktadır.*

**Anahtar Sözcükler:** *Anjioliipoleiomyoma, nadir uterin tümör, pecoma*

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## INTRODUCTION

In recent years, mesenchymal tumors consisting of vascular components, mature adipose tissue, and smooth muscle have been recognized as rare variants of leiomyomas (1). These tumors have been described in the literature under various names, including lipoleiomyomatous tumors, hamartomas, benign lipomatous lesions, and benign mixed mesodermal tumors (1,2). Due to the lack of an official classification by the World Health Organization (WHO), most reported cases in the literature have been classified under angiomyolipoma (AML) or angioleiomyoma (ALM) (1). These tumors have also been referred to in the literature as PEComas, composed of perivascular epithelioid cells, which test positive markers such as desmin, caldesmon, and SMA, and show immunoreactivity particularly with anti-human melanoma (HMB-45) (3,4).

One of the rare tumors of the uterus previously classified under PEComas but recently identified as a distinct entity is angioliipoleiomyoma (ALLM).

ALLM consists of three histological components: abnormal vascular structures, adipose tissue cells, and smooth muscle cells. It is a rare tumor with an incidence rate of 0.06% (5). A literature search was conducted using the keyword "angioliipoleiomyoma" on PubMed and Google Scholar, up to June 2024. Animal studies were excluded from the review. As noted in the introduction, due to the lack of a clear classification, the most recent diagnostic criteria were utilized to identify 31 ALLM cases (Table-1). In our center, two cases of uterine angioliipoleiomyoma were analyzed between 2015 and 2024. One of these cases had been previously reported as a case study by Bacanakgil et al (6).

**Table-1.** Clinical and pathological features of uterine angiolipoleiomyoma cases reported in the literature.

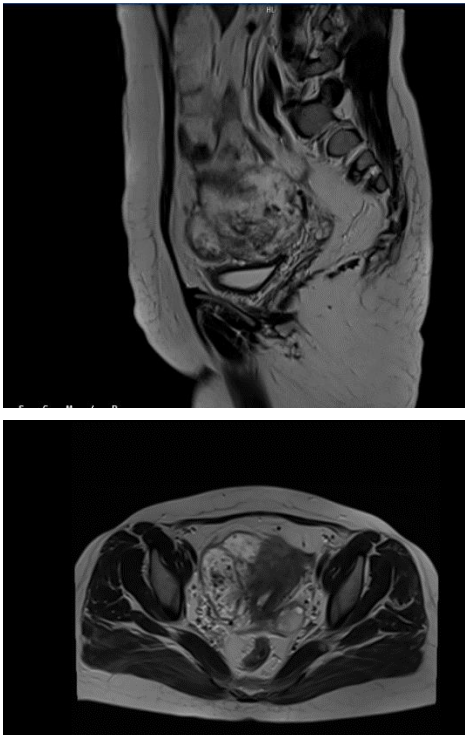
Author	Year	Age	Symptom	Location	Size (cm)	Immunohistochemistry	Final pathology	Other pathology	Management
Jacobs DS et al.	1965	50	Pelvic and lower abdominal pain	Corpus	7	NS	Lipoleiomyoma	Leiomyomas	Hysterectomy
		32	Abnormal uterine bleeding	Corpus	8	NS	Lipoleiomyoma	None	Hysterectomy
		35	Vaginal bleeding	Corpus	7	NS	Lipoleiomyoma	None	Hysterectomy
		77	Lower abdominal pain	Corpus	15	NS	Lipoleiomyoma	Small calcified leiomyomas	Supracervical hysterectomy
		41	Dysmenorrhea and abdominal pain	Corpus	10	NS	Lipoleiomyoma	Endometrial polyp	Hysterectomy
		80	Incidental	Corpus	3	NS	Lipoleiomyoma	Leiomyoma	Necropsy, no operation
		51	NS	Corpus	8	NS	Lipoleiomyoma	Leiomyomas	Hysterectomy
Lo RV et al.	1987	47	Menometrorrhagia	Corpus	5	NS	Lipomatous lesions	NS	NS
Sienski M	1989	52	NS	Corpus	6	NS	Angiolipoleiomyoma	Endometrial hyperplasia	NS
		52	NS	Cervix	16	NS	Angiolipoleiomyoma	None	NS
		57	NS	Cervix	9	NS	Angiolipoleiomyoma	None	NS
Shintaku M	1996	78	Uterine prolapse, Cystocele, rectocele	Corpus, anterior wall	5.3	$\alpha$ -SMA(+) desmin(+) anti-S-100 protein antibody(+)	Angiolipoleiomyoma	Multiple leiomyomas	Vaginal hysterectomy
		67	Uterine prolapse, Cystocele	Corpus	7	$\alpha$ -SMA(+) desmin(+) anti-S-100 protein antibody(+)	Angiolipoleiomyoma	None	Vaginal hysterectomy
		73	Asymptomatic incidental finding at operation for rectal cancer	Corpus, right lateral wall	Unknown	$\alpha$ -SMA(+) desmin(+) anti-S-100 protein antibody(+)	Angiolipoleiomyoma	Rectal cancer, Cervical polyp	Rectal cancer surgery
		74	Uterine prolapse, Cystocele	Cervix	3.5	$\alpha$ -SMA(+) desmin(+) anti-S-100 protein antibody(+)	Angiolipoleiomyoma	Severe dysplasia of the uterine cervix	Vaginal hysterectomy

		60	Lower abdominal distension (due to ovarian tumor)	Cervix	4.2	$\alpha$ -SMA(+) desmin(+) anti-S-100 protein antibody(+)	Angiolipoleiomyoma	Multiple leiomyomas, Mucinous cystadenoma of the left ovary	Hysterectomy
Braun HL et al.	2002	51	Postmenopausal vaginal bleeding	Corpus	2	NS	Angiolipoleiomyoma	None	Hysterectomy
Ren RL	2003	40	Low back and pelvic pain	Corpus	5	$\alpha$ -SMA(+) desmin(+) HMB-45 and melan-a (-)	Angiolipoleiomyoma	Focal atypical leiomyoma	Hysterectomy
Kajo K et al.	2010	53	NS	Corpus	NS	NS	Angiolipoleiomyoma	NS	NS
Bacanakgılı BH et al.	2015	44	Lower abdominal pain	Corpus	7.5	SMA, CD31 and S100 (+), HMB-45 and melan-a (-)	Angiolipoleiomyoma	None	Hysterectomy
Rojas DCP et al.	2016	61	Incidental	Corpus	NS	NS	Angiolipoleiomyoma	Mature cystic teratoma	Hysterectomy
Shakarwal S et al.	2017	28	Lower abdominal pain and spotting	Corpus, left lateral wall	8	NS	Angiolipoleiomyoma	None	Mass excision
Cendek BD et al.	2018	59	Lower abdominal pain	Corpus, posterior wall	6	NS	Angiolipoleiomyoma	None	Hysterectomy
Paryani NS et al.	2020	26	Large abdominopelvic mass	Corpus, posterior wall	12	$\alpha$ -SMA and Calponin Antibody (+)	Angiolipoleiomyoma	None	Open Myomectomy
Garg N et al.	2021	46	Irregular menstrual cycles	Corpus	6	$\alpha$ -SMA(+) desmin(+) HMB-45 (-)	Angiolipoleiomyoma	None	Hysterectomy
Seo G et al.	2022	50	Incidental	Corpus, posterior wall	NS	NS	Angiolipoleiomyoma	NS	Hysterectomy
Psomiadou V et al.	2022	59	Postmenopausal vaginal bleeding	Corpus	5.5	HMB-45 and melan-a (-), Van Gieson and orcein (+)	Angiolipoleiomyoma	None	Hysterectomy
Verocq C et al.	2022	58	Incidental	Corpus	4	Desmin, Caldesmon and CD10 (+), HMB45 (-)	Angiolipoleiomyoma	None	Hysterectomy
Gupta D et al.	2024	40	Chronic lower abdominal pain and abnormal uterine bleeding	Corpus	23	$\alpha$ -SMA(+) HMB-45 (-)	Angiolipoleiomyoma	Mature cystic teratoma	Hysterectomy
Present case	2024	40	Heavy vaginal bleeding and pelvic mass	Corpus, right lateral wall	9	NS	Angiolipoleiomyoma	Leiomyoma	Vaginal myomectomy and Mass excision

## CASE REPORT

A 40-year-old G4P4 patient presented to the emergency department with heavy vaginal bleeding. Except for a history of diabetes mellitus (DM), her medical history was unremarkable. Her hemoglobin level was 6.8 g/dL and hematocrit were 23.9% upon admission. Emergency erythrocyte and fresh frozen plasma transfusion were administered.

Pelvic ultrasound revealed a cystic-solid mass with a hyperechoic, heterogeneous appearance adjacent to the right side of the uterus. A protruding 5 cm active bleeding mass, suspected to be a myoma in status nascent, was excised, followed by dilation and curettage. After stabilizing the patient, an MRI was performed. MRI demonstrated a lobulated mass (90x52 mm) containing mildly hyperintense areas on T1A images, with heterogeneous hyperintensity and punctate enhancement on post-contrast imaging (Figure-1). The mass excised via Pfannenstiel incision, and the final pathology confirmed ALLM.



**Figure-1.** MRI findings of the tumor.

## DISCUSSION

ALLM is usually identified in patients aged 40 years and older (with the exception of one 26-year-old case). It is always benign, sharing some histopathological features with leiomyomas but distinguished by its vascular and adipose components (7).

Typically located in the uterine corpus, these masses range from 2 cm to 16 cm in size, with an average diameter of 7.5 cm. They are often associated with tuberous sclerosis, as 69-80% of tuberous sclerosis patients have concurrent ALLM.

Despite their generally asymptomatic nature, they can occasionally cause irregular menstrual bleeding.

Diagnostic imaging, including ultrasonography, CT, and MRI, may be utilized for preoperative evaluation. However, imaging findings are non-specific (8).

Histologically, immunohistochemistry differentiates ALLM from ALM based on HMB-45 negativity. Verocq et al. reported KRAS and KIT mutations in a case of ALLM in 2022 (9).

## CONCLUSION

The aim of this ALLM case report is to contribute to the classification of benign uterine tumors and enhance diagnostic accuracy by providing clinical insights and imaging characteristics.

**Conflict of interest:** None of the authors have conflict of interest to declare.

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