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CASE REPORT

Uterine Intravenous Leiomyomatosis with Internal Iliac Artery Involvement

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Background: Intravenous leiomyomatosis (IVL) is a rare benign uterine smooth muscle tumor that typically spreads along venous channels. Arterial extension is exceedingly uncommon but presents greater surgical challenges and may adversely affect prognosis.

Case Report: We describe a case of IVL extending into the internal iliac artery in a 32-year-old woman. The diagnosis was established through comprehensive imaging, including ultrasound and magnetic resonance imaging (MRI), and confirmed by histopathology. Given the vascular involvement, the lesion was excised laparoscopically.

Conclusion: In atypical cases of IVL with arterial involvement, surgical complexity is substantially greater than in venous disease, as arterial invasion complicates vascular dissection and hinders complete clearance. This increases the risk of recurrence and may negatively influence long-term prognosis. In the present case, laparoscopic resection allowed precise vascular control, minimized surgical trauma, and facilitated recovery. Early diagnosis, radical excision, and continued surveillance remain essential for optimizing outcomes.

Keywords: intravenous leiomyomatosis, uterine leiomyoma, internal iliac artery, case report

Introduction

Intravenous leiomyomatosis (IVL) is a rare, histologically benign smooth muscle tumor of uterine origin that extends intrapelvically or extrapelvically through the venous system.[1] IVL accounts for less than 0.1% of all uterine leiomyomas,[2] with fewer than 800 cases reported worldwide.[3] Despite its benign histology, IVL may spread through the iliac veins into the inferior vena cava and even the right atrium, creating a risk of sudden cardiac death from impaired cardiac function.[3,4] In a cohort of 361 patients, the overall mortality rate was 18.8%, primarily in advanced cases with cardiac or major vascular involvement.[5] Prognosis is generally favorable after radical resection, with a recurrence rate of approximately 7.6%, whereas preservation of the uterus or ovaries is associated with recurrence rates as high as 75%.[4]

Although IVL usually involves venous structures, rare cases of pulmonary artery extension have been reported.[6–8] Most of these, however, represent arterial compression or secondary invasion from extensive venous tumor burden rather than true intraluminal arterial extension. Color Doppler ultrasonography is useful in the initial assessment of suspected IVL, as it allows visualization of intravascular tumor extension, vascular architecture, and flow dynamics, thereby aiding differential diagnosis and surgical planning.[9,10]

Herein, we report an exceptional case of uterine IVL extending into the internal iliac artery in a 32-year-old woman, diagnosed three months after an uncomplicated delivery and successfully treated with surgical resection. To our knowledge, this is one of the very few documented cases demonstrating true arterial involvement in IVL. This case expands the

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recognized spectrum of IVL-related vascular involvement and highlights the importance of considering the possibility of arterial extension during diagnostic evaluation.

Case Report

A 32-year-old woman, three months after an uncomplicated delivery, was referred to our hospital when a routine follow-up ultrasound at a local clinic incidentally revealed a mixed cystic–solid hypoechoic mass in the right adnexal region. She was asymptomatic and had no specific clinical manifestations. Her medical history included a laparoscopic myomectomy performed two years earlier. She was an occasional smoker, denied alcohol consumption, and had no evidence of obesity. Gynecological examination revealed no abnormalities of the vulva or cervix; however, a distinct mass measuring approximately 5 cm was palpable on the right side of the uterus. All relevant tumor markers, including human chorionic gonadotropin, were within normal limits. Transvaginal ultrasonography demonstrated a mixed echogenic mass measuring 5.53×3.52 × 5.16 cm on the right side of the uterus. The mass was poorly demarcated from the surrounding myometrium and contained areas of hypoechogenicity (Figure 1A). Color Doppler flow imaging (CDFI) revealed abundant intralesional vascularity (Figure 1B). Sonographic elastography showed that the lesion had a soft texture (elasticity score: 2) with indistinct margins, measuring approximately 5.5×3.5 cm (Figure 1C). Contrast-enhanced ultrasound (CEUS) demonstrated irregular, worm-like enhancement at peak contrast (Figure 1D). Quantitative perfusion analysis using time–intensity curves revealed that the lesion enhanced simultaneously with the myometrium but with heterogeneous low enhancement (Figure 1E). Collectively, these findings suggested a uterine origin, with differential diagnoses including IVL or a leiomyoma with degenerative changes. Pelvic magnetic resonance imaging (MRI) further demonstrated an ill-defined right pelvic mass measuring 4.0×6.2 × 4.3 cm (Figure 2A). The lesion showed mixed signal intensity with internal heterogeneity and poorly defined margins. The uterus was displaced to the left, and a thickened, tortuous vascular structure was observed along the anterior uterine wall, suggestive of vascular involvement (Figure 2B).

After excluding contraindications and obtaining informed consent from the patient and her family, laparoscopic resection of the pelvic and uterine lesions was performed. Although hysterectomy was initially recommended given the nature of the disease, the patient—being young and wishing to preserve fertility—strongly requested uterine preservation.

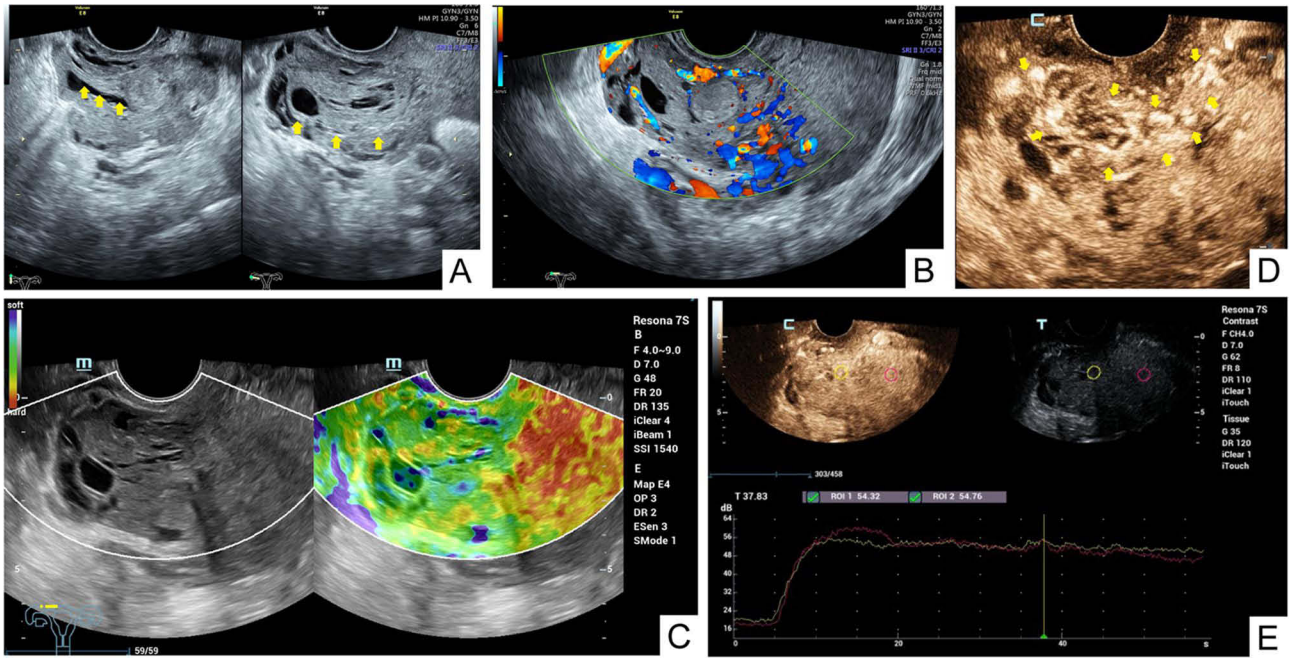

Figure 1 Ultrasound of the pelvic mass. (**A**) Transvaginal ultrasonography revealed a mixed cystic–solid hypoechoic mass in the right adnexal region (arrow). (**B**) Color Doppler flow imaging (CDFI) demonstrated abundant intralesional vascularity. (**C**) Elastography showed that the lesion had a soft texture with indistinct margins. (**D**) Contrast-enhanced ultrasound (CEUS) revealed irregular, worm-like enhancement of the lesion at peak contrast (arrow). (**E**) Time–intensity curve analysis demonstrated simultaneous enhancement with the myometrium and heterogeneous low enhancement.

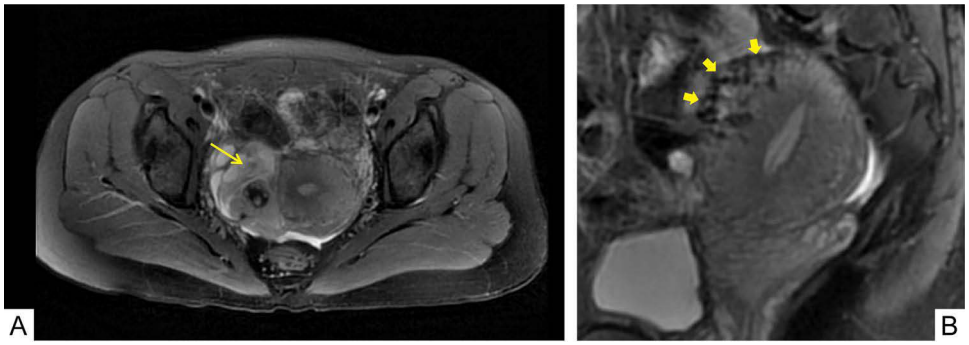

Figure 2 Pelvic magnetic resonance imaging (MRI) findings. (**A**) MRI showed a heterogeneous pelvic mass displacing the uterus leftward. (**B**) A thickened, tortuous vessel was observed along the anterior uterine wall (arrow), suggestive of vascular extension.

Intraoperatively, a leiomyomatous bulge measuring approximately 4×3 cm was identified on the right anterior wall of the uterine body. In addition, an irregularly shaped mass measuring approximately 6×6 × 5 cm was noted on the right paramedian side of the uterus (Figure 3A). On further exposure and dissection, the mass demonstrated cystic and irregular growth patterns extending along the adjacent vasculature, with distal involvement of the internal iliac vein and artery (Figures 3B and C). The lesion was carefully dissected along its margins, and the intravascular thrombus was removed. The affected segments of the right internal iliac vein and artery were resected using an ultrasonic scalpel to achieve complete tumor clearance (Figures 3D and E). Intraoperative frozen-section analysis of the right iliac vascular mass showed proliferating spindle-shaped cells arranged in an interlacing pattern, consistent with a smooth muscle tumor. Final pathological examination confirmed a highly cellular smooth muscle tumor involving the right iliac vessels, with minimal mitotic activity (Figure 4A).

The lesion demonstrated intravascular growth consistent with IVL. Immunohistochemical (IHC) staining showed the following results: B5-01 (2), Ki-67 (15%, +), α-smooth muscle actin (α-SMA, +), desmin (+), CD31 (+, vascular), CD34 (+, vascular), fumarate hydratase (FH, +) (Figure 4B), succinate dehydrogenase subunit B (SDHB, +) (Figure 4C), estrogen receptor (ER, +) (Figure 4D), progesterone receptor (PR, +) (Figure 4E), and phosphohistone H3 (+, isolated cells). Elastin–fibrin staining confirmed tumor invasion into the vascular lumen (Figure 4F), supporting the diagnosis of IVL involving the internal iliac artery.

Postoperatively, the patient received antibiotics and supportive care. Her recovery was uneventful, and she was discharged without complications.

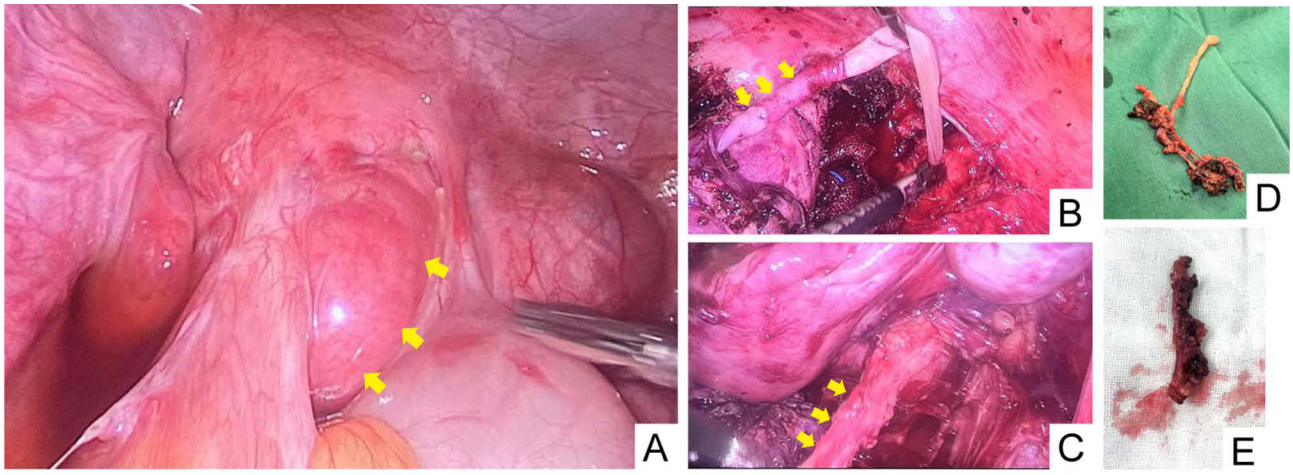

Figure 3 Intraoperative findings and gross specimen morphology. (**A**) Intraoperative view showing a leiomyomatous bulge on the right anterior wall of the uterus (arrow). (**B** and **C**) Dissection revealed an irregular mass extending along adjacent blood vessels, with involvement of the internal iliac vein and artery (arrow). (**D** and **E**) The lesion was excised along its margins; intravascular thrombus was removed, and the affected segments of the internal iliac vessels were resected using an ultrasonic scalpel.

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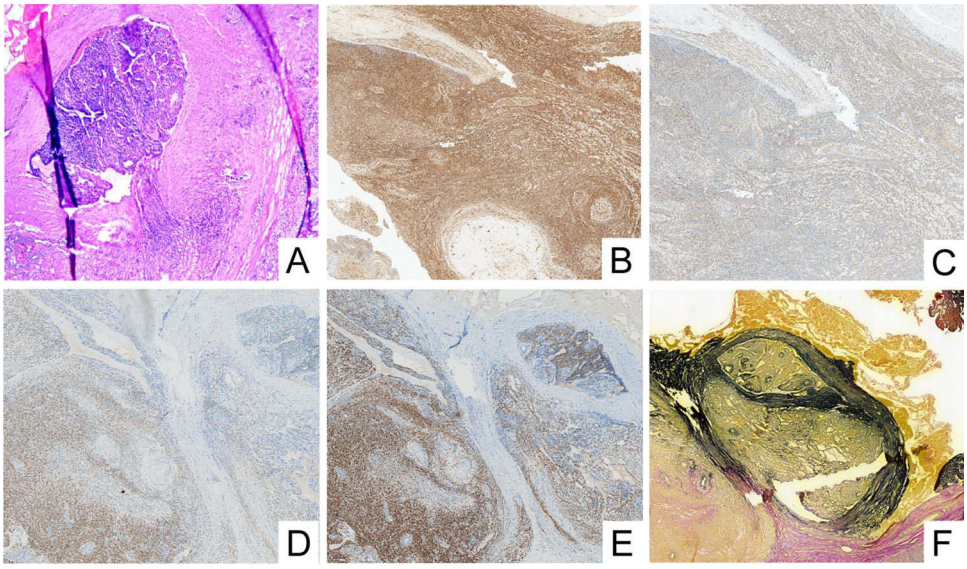

Figure 4 Histopathological and immunohistochemical (IHC) features of the lesion. (**A**) Hematoxylin and eosin staining showed a highly cellular smooth muscle tumor with minimal mitotic activity. (**B**) Fumarate hydratase (FH) positivity (magnification, ×200). (**C**) Succinate dehydrogenase subunit B (SDHB) positivity (magnification, ×200). (**D**) Estrogen receptor (ER) positivity (magnification, ×200). (**E**) Progesterone receptor (PR) positivity (magnification, ×200). (**F**) Elastin–fibrin staining confirmed tumor invasion into the vascular lumen (magnification, ×200).

Discussion

IVL is a rare benign smooth muscle tumor of uterine origin, characterized by intravascular growth along venous channels and, in exceptional cases, extension into the cardiac chambers.[4,11] Epidemiological studies indicate that IVL accounts for less than 0.1% of all uterine leiomyomas, with fewer than 800 cases reported worldwide,[2,3] underscoring both its rarity and the importance of documenting atypical presentations. Here, we describe an exceedingly rare case of uterine IVL with arterial involvement, extending into the internal iliac artery of a 32-year-old woman diagnosed three months postpartum. The diagnosis was established through comprehensive imaging and pathological evaluation, and the lesion was successfully treated with laparoscopic resection. This case expands the recognized spectrum of vascular involvement in IVL and highlights the need to consider atypical arterial extension during diagnostic evaluation.

Although the exact pathogenesis of IVL remains unclear, two main theories have been proposed. One suggests that IVL develops from uterine leiomyomas, with tumor cells infiltrating and proliferating within venous channels.[3,4] The alternative hypothesis posits that IVL originates from smooth muscle cells within the walls of pelvic veins that undergo neoplastic transformation.[12] IVL typically spreads along venous pathways, and involvement of the internal iliac artery is exceedingly rare. Several mechanisms may account for this phenomenon. One possibility is that pelvic arteriovenous anastomoses provide a conduit for tumor cells to migrate from the venous system into adjacent arteries.[7] Another explanation is direct invasion through the venous wall into the arterial lumen, particularly in regions of prior vascular injury or remodeling, such as those following surgery.[13] In the present case, the history of laparoscopic myomectomy may have contributed to local vascular alterations, facilitating tumor infiltration into the arterial system. In addition, increased pelvic blood flow and hormonal changes associated with pregnancy and the postpartum period may have further promoted tumor progression and vascular dissemination.

We used elastography to evaluate the lesion; however, its diagnostic value for IVL is limited. To improve diagnostic accuracy, we additionally performed CEUS with time–intensity curve analysis for preliminary differentiation from uterine sarcoma. Uterine sarcomas typically demonstrate rapid early-phase enhancement followed by diffuse, heterogeneous peak enhancement on CEUS.[14] In contrast, the IVL lesion in our patient exhibited uniform low enhancement throughout the tumor, providing greater diagnostic specificity than color Doppler, which only reveals increased vascularity. By enabling real-time assessment of microvascular perfusion and quantification of contrast enhancement kinetics, CEUS offers diagnostic information beyond that of Doppler imaging, thereby increasing confidence in distinguishing IVL from uterine sarcoma.[3]

A major clinical challenge is the misdiagnosis of uterine leiomyomatosis as a malignant uterine neoplasm, particularly uterine sarcoma. Uterine sarcomas often exhibit nonspecific imaging features—such as irregular margins, heterogeneous echogenicity, cystic degeneration, and increased vascularity—that may overlap with those of benign leiomyomas or leiomyomatosis.[14] As a result, many benign cases are identified only after hysterectomy or myomectomy performed under a presumptive diagnosis of sarcoma.[15] Such misdiagnoses can lead to overtreatment, including unnecessary radical procedures.[15] Accurate preoperative differentiation therefore requires integration of detailed clinical history, multimodal imaging, and, when feasible, intraoperative frozen-section analysis to minimize the risk of inappropriate surgical intervention.

In this case, histological analysis revealed a tumor with high cellular density but minimal mitotic activity. Importantly, no cytological atypia was observed; the nuclei were relatively uniform, without diffuse or marked pleomorphism, and no evidence of coagulative necrosis was identified. According to current risk stratification criteria, pronounced atypia and coagulative necrosis are essential for diagnosing leiomyosarcoma, whereas their absence or only focal presence suggests a smooth muscle tumor of uncertain malignant potential (STUMP).[16] The tumor also demonstrated diffuse ER and PR positivity, indicating hormonal responsiveness—a characteristic feature of benign smooth muscle tumors but rarely seen in leiomyosarcomas.[4] Elastin–fibrin staining confirmed tumor cells within the true vascular lumen, excluding perivascular infiltration.[2] Physiological changes during pregnancy may have contributed to the observed tumor behavior. Elevated estrogen and progesterone levels can stimulate tumor growth and facilitate intravascular extension, while pregnancy-related hemodynamic alterations, including increased uterine blood flow and vascular remodeling, may create a permissive microenvironment for invasion into both venous and arterial lumens.[3] These factors likely explain arterial involvement in young postpartum patients and align with previous reports that IVL exhibits hormone-dependent growth and often arises or progresses during or shortly after pregnancy.[3] Collectively, these findings support the diagnosis of IVL with arterial extension while effectively excluding leiomyosarcoma and STUMP.

Molecular analyses have shown that IVL harbors genetic alterations distinct from those of conventional uterine leiomyomas. In particular, mutations in exon 2 of the MED12 gene—commonly detected in typical leiomyomas—are rarely identified in IVL, suggesting a divergent pathogenic pathway.[3] In addition, deficiencies in other markers detected by IHC have been associated with IVL and may indicate an underlying hereditary tumor syndrome.[2,5] Supplemental molecular analyses are therefore recommended in young patients, in individuals with multifocal or recurrent lesions, and in cases with atypical morphology, as these findings can guide genetic counseling, systemic surveillance, and personalized follow-up strategies. Molecular analysis has not yet been performed for the lesion in this patient.

Surgical resection remains the primary treatment for IVL, with complete tumor excision essential to minimize recurrence. For disease confined to the pelvic veins, hysterectomy with bilateral salpingo-oophorectomy is considered the standard approach.[17] A meta-analysis showed that laparoscopic myomectomy reduces blood loss, shortens hospitalization, and lowers analgesic requirements compared with open surgery, without increasing complications.[18] Reproductive outcomes, including pregnancy rates and obstetric endpoints, were comparable between approaches, and no cases of uterine rupture were reported.[18] Although minimally invasive surgery may involve higher direct costs, its benefits of faster recovery and earlier return to work may reduce overall societal costs.[18] In cases with cardiac extension, a staged or combined gynecologic and cardiovascular surgical strategy is often required.[19] For patients with extensive vascular involvement or pelvic vascular anomalies, preoperative embolization of feeding vessels has been described as an adjunct to reduce intraoperative bleeding and facilitate dissection. Nonetheless, embolization alone is insufficient and cannot substitute for complete resection, which remains the only definitive curative treatment.[20]

Compared with typical venous involvement, arterial extension in IVL may represent a more aggressive clinical phenotype. Previous reports suggest that arterial invasion reflects a greater capacity for intravascular dissemination and is associated with increased surgical complexity, making complete resection more challenging.[4,7,21] Evidence from recent case reports further illustrates the heterogeneity of vascular involvement, surgical strategies, and clinical outcomes in IVL (Table 1). Because incomplete resection remains the primary risk factor for recurrence, arterial involvement may indirectly increase recurrence risk by complicating surgical clearance.[13,17] Moreover, even apparently complete resections can be followed by recurrence attributable to residual tumor or micrometastases, highlighting the need for long-term surveillance with serial imaging.[22] Hormonal therapy has been explored in patients with residual disease, although its efficacy remains inconclusive.[23] Accordingly, patients with arterial extension should undergo closer follow-up with contrast-enhanced computed tomography

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Table 1 Summary of Reported Intravenous Leiomyomatosis Cases with Vascular Involvement (2015–2025)

First Author (Year)	Involved Vascular Structure	Imaging Evidence	Surgical Approach	Arterial Invasion	Follow-up	Recurrence
Jiang, 2025[24]	IVC → right atrium	Ultrasound, Echo, CT, MRI	Multidisciplinary complete resection	No	1 month follow-up	No
Dong, 2024[25]	Right ovarian vein → right internal iliac → IVC → right atrium	[^{18}F] F-FAPI PET/CT	NR	No	NR	NR
Bahlouli, 2024[26]	IVC → Right atrial	Echo, CT	Combined gynecologic and cardiac surgery; resection of tumor involving IVC and right atrial	No	A few months follow-up	Yes, re-operation
Gonzalez-Urquijo, 2024[27]	Intracardiac IVL (3 cases)	Echo, CT, MRI (multimodality)	Sternotomy and laparotomy with extracorporeal circulation; tumor removal and IVC/iliac vein ligation	Yes (pulmonary artery)	10-, 13-, and 37-year follow-up	No
Huang, 2023[7]	Extension to pulmonary artery; right heart involvement	Echo, CT, CTA (multimodality)	Staged tumor resection (cardiothoracic then gynecologic)	Yes (pulmonary artery)	3 months follow-up	No
Aleksandrov, 2023[28]	Pelvic veins → IVC → right heart	US, CT, MRI, echo	Multidisciplinary resection (cardiac and pelvic)	No	1 year follow-up	No
Garcés-Garcés, 2023[29]	Pelvic veins → IVC → right atrium	Echo, CT, MRI	Complete removal	No	2 years follow-up	No
Kan, 2022[20]	Pelvic arteriovenous fistula with IVL	CTA, DSA	TAH-BSO and IVL excision; some with adjunct embolization	No	4–6 months follow-up	Yes
Shaked, 2022[30]	IVC →right heart chambers	Echo, CT	Sternotomy and laparotomy; IVL resection	No	1 month	No
Akinseye, 2020[8]	Femoral vein → IVC → right atrium → pulmonary artery	Ultrasound, Echo, CT	Combined resection under CPB; gynecologic tumor control	Yes (pulmonary artery)	NR	NR
Zeng, 2017[31]	Pelvic veins → bilateral iliac veins → bilateral renal veins → IVC → right atrium	Ultrasound, CT, Echo	One-stage combined resection (pelvis, IVC, renal veins and right atrium), right oophorectomy	No	Ongoing follow-up	No
Thompson, 2016[32]	Left ovarian vein → renal vein → IVC → right atrium and internal iliac branches	CT	TAH-BSO and sternotomy; CPB with multiple cavotomies, tumour removal	No	Reported; good postoperative recovery	NR
Jain, 2015[33]	Left ovarian vein → left renal vein → IVC →right atrium →right ventricle	Ultrasound, CT, MRI	Thoracotomy with right atrium /IVC tumour removal, then TAH-BSO	No	Reported; no residual lesion	No

Abbreviations: IVC, Inferior Vena Cava; Echo, echocardiography; CT, computed tomography; MRI, magnetic resonance imaging; PET/CT, positron emission tomography/computed tomography; NR, not reported; IVL, intravenous leiomyomatosis; CTA, computed tomography angiography; DSA, digital subtraction angiography; TAH-BSO, total abdominal hysterectomy and bilateral salpingo-oophorectomy; CPB, cardiopulmonary bypass.

or MRI at shorter intervals—every 3–6 months during the first two years, followed by annual imaging thereafter—to allow timely detection of tumor regrowth or delayed dissemination.[5,22]

Conclusion

In conclusion, IVL requires a multidisciplinary approach for optimal management, particularly when atypical vascular involvement is present. Early diagnosis, complete surgical excision, and rigorous postoperative surveillance are essential to achieving favorable outcomes and minimizing recurrence. Although IVL usually arises from venous structures, the rare arterial extension observed in this case increases surgical complexity and necessitates long-term monitoring. This case highlights an important clinical lesson: awareness of such atypical presentations is critical for guiding treatment strategies and improving patient prognosis.

Ethical Approval

Ethical approval was obtained from the Human Research Ethics Committee of the Second Affiliated Hospital, School of Medicine, Zhejiang University.

Informed Consent

Written informed consent was obtained from the patient for participation in this case study and for publication of clinical data and images.

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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.

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Disclosure

The authors declare no competing interests in this work.

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