



RESEARCH ARTICLE

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CAVERNOUS HEMANGIOMA IN UTERINE LEIOMYOMA: A HEALTH SYSTEM RESPONSE TO A RARE FERTILITY CHALLENGE

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ABSTRACT

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Rare gynecological pathologies can closely mimic more common conditions, leading to delayed diagnosis and treatment. We report the case of a 37-year-old woman with a decade-long history of secondary infertility who presented with a large uterine mass. Radiology suggested a fibroid with cystic degeneration, but histopathology revealed a cavernous hemangioma within an intramural leiomyoma, confirmed by CD34 immunostaining. This case illustrates that histopathology remains the gold standard when radiological findings are atypical, and highlights the importance of maintaining diagnostic suspicion in unusual presentations. Fertility-preserving decisions in such cases require careful balancing of surgical risks with patient priorities and clear preoperative counselling. At a broader healthcare level, building stronger diagnostic pathways, such as incorporating angiography and advanced imaging could help doctors recognize rare uterine conditions before surgery, leading to safer and more tailored care for patients. Finally, the patient's favourable outcome despite complications underscores the critical role of multidisciplinary teamwork in managing rare and complex cases.

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INTRODUCTION

Hemangiomas are benign tumors that originate from the endothelial cells of blood vessels, or from the pericytes on the blood vessel wall. [1] In the uterine corpus, they commonly involve the myometrium diffusely. [2] Majority of the hemangiomas are incidental findings, because of their asymptomatic nature and small size. However, they can present as gynecological and obstetric complications, ranging from intermenstrual spotting, menometrorrhagia, and infertility to maternal and fetal death from excessive bleeding of the gravid uterus. [3]

ORGANIZATIONAL CONTEXT

This case was managed at a tertiary care teaching hospital in India that serves as a referral center for complex gynecological cases. The hospital has access to diagnostic modalities such as ultrasonography, MRI, and immunohistochemistry, and it provides both conservative and surgical management for uterine pathology.

Despite these resources, there is a lack of established protocols for diagnosing and managing extremely rare uterine tumors. The team had to rely on individual expertise and multidisciplinary collaboration between gynecologists, pathologists, radiologists, anesthesiologists, and critical care specialists. The case also unfolded within a broader health system where infertility carries significant psychosocial implications. The patient's priority was fertility preservation, which influenced the clinical decision-making process.

PERSONAL CONTENT

The patient was a 37-year-old woman with a decade-long history of secondary infertility. She had experienced a spontaneous abortion 13 years earlier and had been unable to conceive after that. Over the preceding two months, she developed an enlarging abdominal mass that caused significant anxiety and physical discomfort. From her perspective, the main concerns were her inability to conceive, her deteriorating health, and the uncertainty of diagnosis. She expressed a strong preference for fertility-preserving treatment.

For the clinical team, the challenge was not only to manage her symptoms and mass but also to respect her reproductive wishes. The surgeons had to weigh the risks of hemorrhage and complications against the potential to conserve her uterus.

PROBLEM

The central problem was diagnostic ambiguity.

Preoperative investigations, including ultrasonography and MRI, showed a large uterine mass consistent with leiomyoma with cystic degeneration (Fig 1a). There was no radiological suspicion of an underlying vascular lesion.

This meant that:

- The surgical team had no opportunity to prepare for a potential vascular tumor.
- There was a risk of massive intraoperative bleeding, which could have been fatal.
- The patient’s anemia complicated surgical planning.
- There were no standard protocols or guidelines to follow for this rare presentation.

This case illustrates a broader health system problem: rare diseases are often diagnosed only after surgery or histopathology, leaving clinicians to manage them without adequate preparation.

SOLUTION

The decision was made to proceed with exploratory laparotomy and myomectomy, in line with the patient’s preference for fertility preservation.

Operative Findings: The uterus was enlarged, and a large intramural fibroid was identified. There was unpredicted excessive bleeding during excision, which the surgical team managed to control. Her preoperative hemoglobin levels of 9.3g/dl had decreased subsequently to 7.1g/dl. She was transfused with two bags of packed cells. The excised specimen measured 16 × 10 × 9 cm. Gross examination revealed an area with dilated blood vessels and clots (Fig 1b).

Histopathology and Immunohistochemistry: Microscopic examination showed spindle cells arranged in fascicles, consistent with leiomyoma, but also revealed dilated cavernous vascular spaces filled with red blood cells and thrombi (Fig 1c). The endothelial lining was bland and flat. Immunohistochemistry demonstrated CD34 positivity, confirming vascular origin and establishing the diagnosis of cavernous hemangioma within leiomyoma (Fig 1d).

Postoperative Course: The patient’s postoperative recovery was complicated by dengue infection and respiratory failure, both of which required intensive supportive care.

Table 1. Comparison with other case reports

Characteristic	Nishikawa et al ^[6]	Celik et al ^[7]	Present study
Case reported on	2022	2017	2023
Age	54	38	37
Symptom	Vaginal bleeding	Vaginal bleeding	Secondary infertility
Radiological diagnosis	Lipoleiomyoma	Leiomyoma with cystic component	Leiomyoma with cystic myxomatous degeneration
Procedure	Hysterectomy	Hysterectomy	Myomectomy
Gross findings	Multiple intramural fibroids	Intramural nodule with blood filled spaces	Large fibroid with dilated vessels showing thrombi
Size of lesion	140x100mm	55x50mm	105x103mm
Location	Myometrium	Myometrium	Myometrium
Histopathological diagnosis	Uterine lipoleiomyoma with hemangioma	Cavernous hemangioma arising in an intramural leiomyoma	Cavernous hemangioma in an intramural leiomyoma
IHC	CD 31 positive	CD 31 positive	CD 34 positive

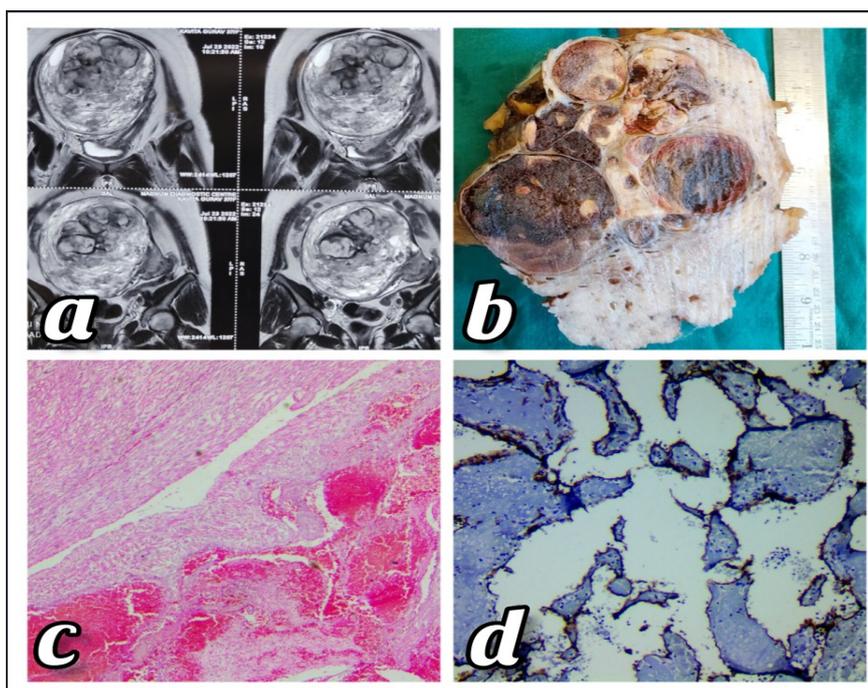


Figure 1. a) MRI abdomen- showing bulky uterus with leiomyoma. b) Gross specimen of Leiomyoma showing hemangioma with thrombosed vessels. c) Leiomyoma with dilated vascular spaces filled with blood (H&E, x40). d) Endothelial cells showing CD 34 positivity (IHC, x40)

With multidisciplinary management, she recovered and was discharged on postoperative day 10. At one-month follow-up, she was asymptomatic and reported resolution of her abdominal discomfort. While her long-term fertility outcomes remain unknown, the surgery achieved both symptom control and preservation of reproductive potential.

DISCUSSION

Uterine hemangioma was first described in 1897 as an incidental discovery during the autopsy of a maternal death.^[5] The exact incidence remains unclear owing to the smaller number of cases reported. Based on the review of literature, less than 60 cases have been reported to date. Among them, uterine hemangioma with concurrent leiomyoma has been reported only twice.^[6,7] On comparative study with other case reports (Table 1), it was found that the lesion was seen in middle-aged woman, presenting as vaginal bleeding or secondary infertility. These leiomyomas were seen to have cystic component in radiology, which should raise the suspicion of hemangioma. All the cases were conservatively managed, owing to the clinico-radiological diagnosis of leiomyoma alone.

And the presence of hemangioma was revealed only after the final histopathological report which was confirmed by immunoreactivity to vascular endothelial markers like CD 34 or CD 31. We had considered the differential diagnosis of angioleiomyoma, a rare variant of leiomyoma, seen in middle aged woman.^[8] Severe dysmenorrhea is its main symptom^[9], which was absent in our case. Microscopically, they are characterized by interlacing short spindle cells along with abundant thick-walled muscular vessels distributed diffusely.^[9] The possibility of angioleiomyoma was eliminated, due to the inconsistent histopathological features. The routine investigations like speculum examination, endometrial curettage and ultrasound have proven to be inconclusive for uterine hemangioma. If there is strong clinical suspicion, a pelvic angiogram and CT may help in confirming the lesion.^[2]

Treatment of uterine leiomyoma depends on its size and whether to preserve the fertility of the patient. Usually, smaller ones are managed conservatively, whereas, larger ones are treated by myomectomy or hysterectomy. On the contrary, uterine hemangiomas are managed by electrocauterization, uterine artery embolization, local excision, laser ablation, internal artery ligation, etc.^[2] Hysterectomy is done, in refractory cases only. In our case, open myomectomy was done, for the removal of leiomyoma, as concurrent uterine hemangioma was not suspected.

Uterine hemangioma was not determined during initial investigations of our patient. This led to delay in treatment, secondary infertility and progressive anemia. There are case reports where inaccurate preoperative diagnosis has led to life-threatening complications due to massive bleeding during uterine interventions.^[3] Fortunately, the surgical procedure was well tolerated by our patient. In summary, the possibility of uterine hemangioma must be considered in cases of abnormal uterine bleeding/ secondary infertility, especially, if radiologically there is leiomyoma with cystic component. Pelvic angiogram may be advised. Arterial embolization or ligation should be considered wherever necessary.

UNRESOLVED QUESTIONS

- Can advanced imaging reliably distinguish leiomyoma with cystic degeneration from leiomyoma with hemangioma?
- What are the long-term fertility outcomes in women treated conservatively for this condition?
- Should high-risk patients be referred to centers with angiography and interventional radiology capacity before surgery?

LESSONS FOR THE FIELD

1. Comparison with earlier case reports demonstrates that such lesions are typically identified in middle-aged women presenting with infertility or abnormal bleeding. Radiological suspicion often suggests leiomyoma with cystic changes, while the true diagnosis emerges only on histopathology (Table 1).
2. Consider rare pathologies in atypical cases. Leiomyomas with cystic or unusual radiological features may harbor unexpected pathology. Clinicians should maintain diagnostic curiosity and suspicion.
3. Diagnostic pathways need strengthening. Standard investigations (ultrasound, MRI, curettage) are often inconclusive for vascular lesions. Pelvic angiography or CT angiography could improve preoperative detection.
4. Patient-centered decision-making is critical. Fertility preservation was a priority for this patient and guided the surgical approach, even in the face of uncertainty. Shared decision-making helped align treatment with patient values.
5. Multidisciplinary care saves lives. The postoperative complications demonstrated the importance of coordinated care among gynecology, pathology, radiology, anesthesiology, and critical care teams.
6. More research and guidelines are needed. Rare conditions such as cavernous hemangioma in leiomyoma are poorly documented, leaving clinicians without standardized protocols. Health systems should encourage case reporting and literature reviews to guide future care.

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GLOSSARY OF ABBREVIATIONS

- CT - Computed Tomography
- H&E -Hematoxylin and Eosin
- IHC - Immunohistochemistry
- MRI - Magnetic Resonance Imaging
- CD - Cluster of Differentiation

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