Asia Pacific Science Press

Asia Pacific Journal of Clinical Medical Research
ISSN(O):3079-8345
ISSN(P):3079-8337
Vol. 2 No.2 (2026)

A Giant Uterine Hydropic Leiomyoma with Distinctive Radiological Patterns: A Challenging Case and Literature Review

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Abstract: Uterine hydropic leiomyoma is a rare condition. Only a few relevant cases have been reported until now. Distinguishing between uterine hydropic leiomyoma and other uterine malignancy tumors, such as leiomyosarcoma, using radiological-based methods is crucial. This can provide vital information on the inherent characteristics, facilitating clinical decision-making. We present a case report of a 42-year-old female patient diagnosed with a giant uterine hydropic leiomyoma with unique radiological characteristics. The patient recovered safely and remained in good condition after a successful surgical intervention.

Keywords: Hydropic Leiomyoma; Hydropic Degeneration; Radiological; Literature Review
Published: Apr 7, 2026
DOI: https://doi.org/10.62177/apjcmr.v2i2.1222

1. Introduction

Leiomyomas are the most prevalent benign mesenchymal neoplasms of the uterus, with an incidence rate of 2.5%. It occurs in approximately 70% of women of menopause age, with heavy menstrual bleeding in approximately one-third of these patients [1,2]. Leiomyoma of the uterus originates from the mesenchymal tissues and is considered to be a monoclonal tumor of smooth muscle cells (SMCs) in the uterus [3,4]. The degeneration of leiomyomas may present different histological characteristics. Leiomyomas can be classified into several subtypes based on their pathology [5]. Hydropic leiomyoma, with a prominent edematous stroma causing compartmentalization of the smooth muscles, is extremely rare [5]. Herein, we report a case of uterine hydropic leiomyoma.

2. Case presentation

A 42-year-old woman with regular menstruation visited a community hospital, complaining of progressive abdominal pain over the past 12 months. Clinical evaluation revealed postpartum perineum, slight leucorrhea, cervical hypertrophy, mild pain, and a bulky mass with poor mobility. Laboratory results revealed mild anemia with a hemoglobin level of 112 g/L (normal range: 115–150 g/L) and a hematocrit of 33.2% (normal range: 35–45%). Other laboratory tests and tumor markers were within normal limits.

Abdominal ultrasonography indicated a well-demarcated heterogeneous echogenic mass in the abdominopelvic region, measuring 18.1 × 12.6 × 8.3 cm, extending to the navel, with honeycomb-like septations inside and dotted blood flow signals around it (Figure 1). For subsequent evaluation, the patient was transferred to our hospital. Abdominopelvic non-contrast-enhanced computed tomography (CT) scan revealed a heterogeneous low-density cystic mass with thickened septations. To determine the tumor's origin, contrast-enhanced CT and routine magnetic resonance imaging (MRI) were performed successively. The contrast-enhanced CT showed intensified heterogeneous thickened septations with a degree of mild to moderate, while the mass's low-density zones showed no enhancement (Figure 2), MRI revealed a flat, elastic cystic component with a complex signal, including a heterogeneous low T1 signal, mixed high T2 signal, and atypical flow voids. DWI showed a hypointense signal (Figure 3).

Figure 1. A well-demarcated heterogeneous echogenicity mass in the abdominopelvic, honeycomb-like septations inside (A), and the dotted blood-flow signal around (B).

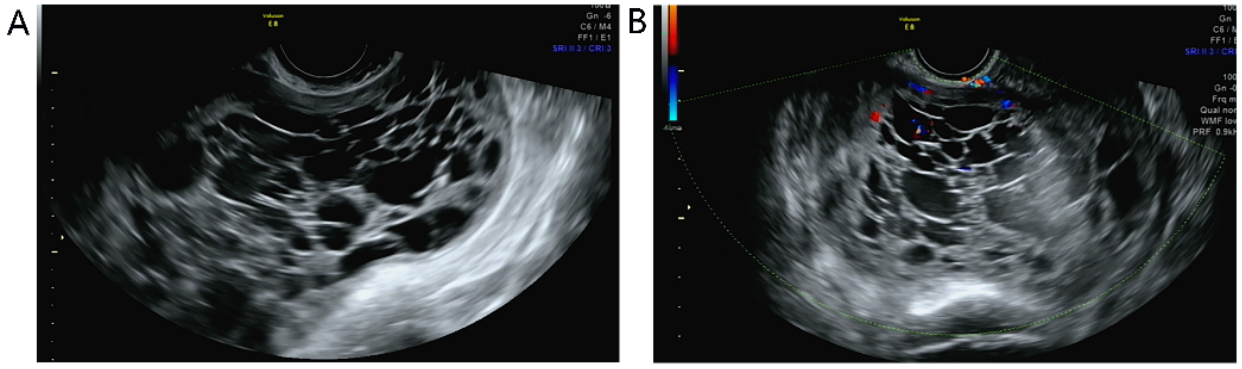


Figure 2. The heterogeneous thickened septations were intensified to a mild-to-moderate degree, while the low-density zone of the mass showed an absence of enhancement on contrast-enhanced CT (A, arterial phase B, venous phase C, equilibrium phase D, non-contrast-enhanced CT).

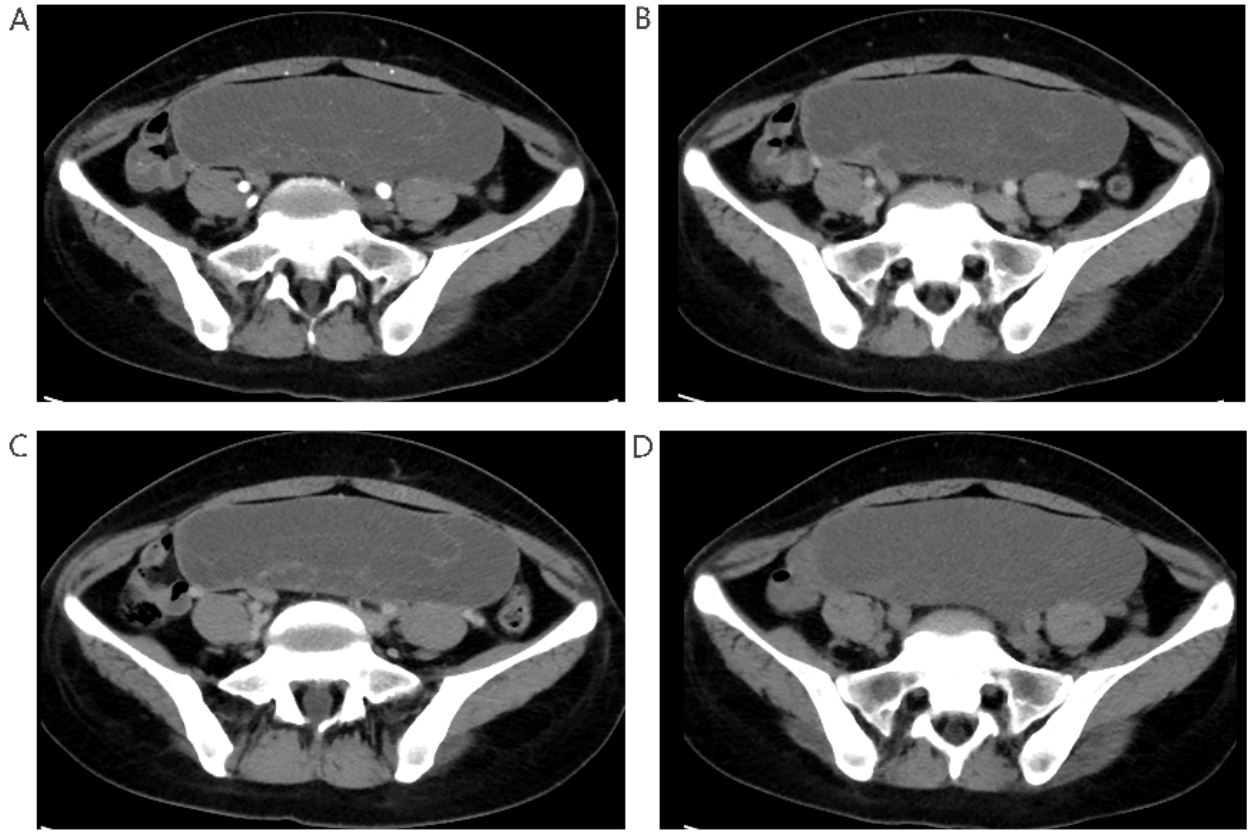
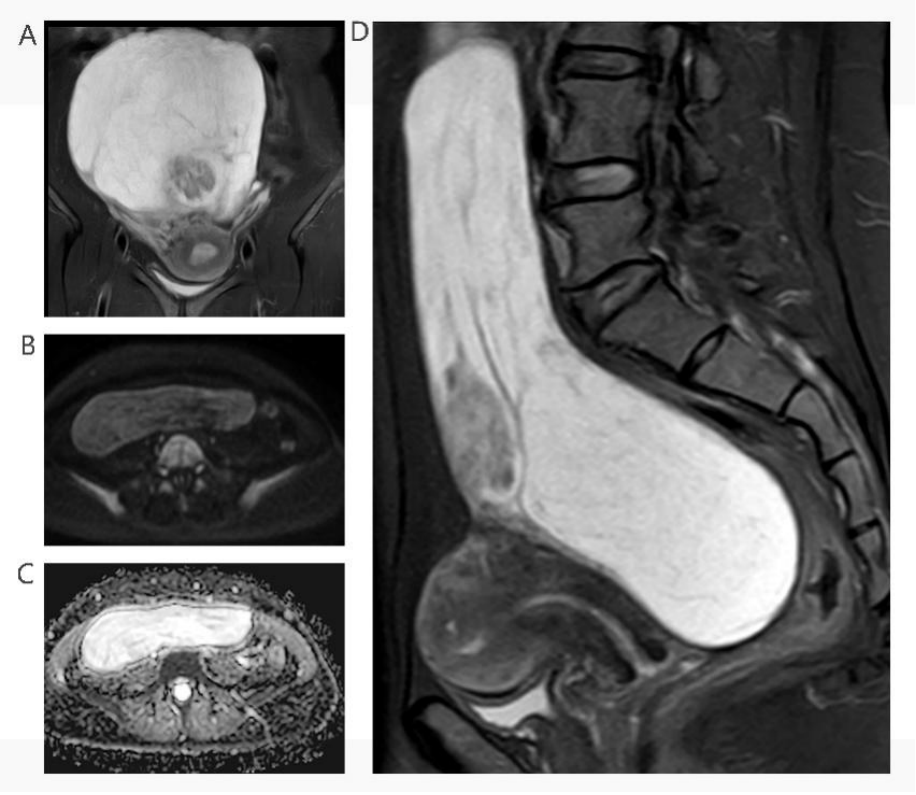
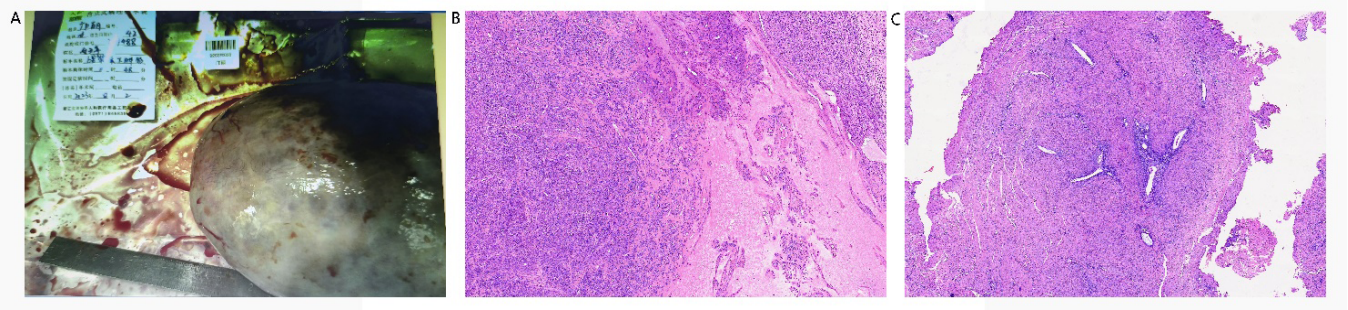


Figure 3. A&D, coronal, sagittal, T2 SPIR, a mixed high T2 signal combined with atypical flow voids. B&C, a hypointense DWI with a hyperintense ADC.



A benign neoplastic lesion was suspected, leading to surgical intervention. A subserosal tumor, measuring approximately 18 × 15 × 10 cm, was observed on the posterior wall of the uterus. It was cystic, soft, containing yellowish fluid, and attached by a thin stalk of 3 cm. Intraoperative pathology suggested a hydropic leiomyoma. Additionally, an interparietal myoma near the right uterine horn measuring 1.5 × 1.5 cm, was found (Figure 4). The patient safely returned to the ward post-surgery and remained in good condition during the 6-month follow-up period.

Figure 4. Macropathology depicts a grayish-white mass, measuring 18.1 × 12.6 × 8.3 cm (A); the tumor cells are separated by excessive amounts of extracellular material (HE staining, ×4) and (B) an interparietal myoma was observed near the right uterine horn, measuring 1.5 × 1.5 cm (D).



3. Discussion

Uterine leiomyoma, an important cause of morbidity among women of reproductive age, is marked by disordered smooth muscle proliferation, changed ECM deposition, and increased hormonal responsiveness, often leading to heavy menstrual bleeding and iron-deficiency anemia [6–8]. Based on the 5th edition of the female genital tumor classification by World Health Organization, the subtypes of leiomyoma include leiomyoma with bizarre nuclei, cellular leiomyoma, fumarate hydratase-deficient leiomyoma, mitotically active leiomyoma, hydropic leiomyoma, apoplectic leiomyoma, lipoleiomyoma, epithelioid leiomyoma, myxoid leiomyoma, dissecting leiomyoma, and diffuse leiomyomatosis [5].

Hydropic leiomyoma, an uncommon yet intriguing variant of uterine leiomyoma, is characterized by zonally segregated

edema, hypervascularity, and the arrangement of tumor cells in nodules or cords, along with distinctive features on medical imaging [9]. We searched PubMed for "hydropic leiomyoma" and "hydropic degeneration" and retrieved 22 records, which included 21 case reports and 1 research paper. Among these, 12 focused on imaging findings, 5 on pathology, and 1 on chromosome variation. Several older articles provided only descriptive reports without detailed imaging and pathology analysis. Typically, ultrasound imaging reveals a multilocular cystic area with hypoechogenicity, whereas color Doppler shows moderate vascularity near the uterine attachment and sparse vascularization peripherally [10–14, 18–19]. In the present study, the decreased blood flow and significant multilocular cystic areas within the lesion suggest a more complete process of degeneration and a less aggressive nature. We identified a cystic-dominated mass with heterogeneous thickened septations on CT and MRI imaging, exhibiting a more typical radiological pattern compared to previous literature [14–18, 28–30]. Imaging characteristics revealed both macro- and microscopic tumor characteristics, with abundant watery edema exhibiting T2 hyperintensity, whereas the tumor's thick-walled vessels appear as cord-like structures with T2 hypointensity [15, 16]. Microscopically, the accumulation of watery edema fluid is usually associated with hyalinized blood vessels and collagen deposition [17]. Griffin B. B. et al. reported that hydropic leiomyoma, distinguished by a large tumor size with edematous tumor cells exhibiting round-oval nuclei arranged in cords or perinodular patterns around vessels and increased thick-walled vessels, varies from usual leiomyoma. Immunohistochemistry showed high mobility group AT-hook 2 (HMGA2) overexpression and rearrangement [9]. Along with hydropic leiomyoma, similar imaging findings can be observed in uterine fibroids with cystic and myxoid degeneration. In uterine fibroids with cystic degeneration, considerable edematous changes can lead to the complete replacement of neoplastic cells. MRI imaging typically shows T2 hyperintensity signals without enhancement on post-contrast sequences. In the case of uterine fibroids with myxoid degeneration, MRI typically reveals T1 hypo- or hyperintensity, T2 hyperintensity, and DWI hyperintensity signals. This is due to the accumulation of mucopolysaccharides and proteins, with smooth muscle cells separated by hyaluronic acid-rich mucoid substances, giving the tissue a soft texture, transparency, and well-defined solidity [19, 20]. Nevertheless, the imaging findings of uterine leiomyoma with myxoid degeneration can resemble those of myxoid leiomyosarcoma, a sarcoma with poor outcomes characterized by large spindle cells of smooth muscles, which may complicate clinical diagnosis [21–23]. Furthermore, smooth muscle tumors of uncertain malignant potential, endometrial stromal sarcomas, adenosarcoma, and uterine carcinosarcoma can mimic the cystic degeneration of uterine leiomyoma and exhibit similar radiological patterns due to changing degrees of cystic degeneration and necrosis, which can contribute to the same radiological patterns [24]. Radiological methods can be challenging in distinguishing these diseases; however, there are still diagnostic clues to follow. Usually, uterine malignant tumors are more aggressive, characterized by rich blood supply, rapid growth, rapid early enhancement, central necrosis, and ill-defined, irregular margins. However, benign tumors are more indolent, with poor blood supply, slow growth, mild or delayed enhancement, and well-defined margins [25–27]. Studies have reported that lobulated borders, T2 dark areas, necrosis, hyperintensity of the tumor compared with the myometrium post-contrast administration, "split fiber" sign with limited or poor enhancement post-contrast, and a high signal on b1000 DWI can differentiate between atypical leiomyomas and leiomyosarcomas [14, 31].

4. Conclusion

To summarize, we report a giant uterine hydropic leiomyoma with unique radiological patterns. Our findings can improve the understanding of its clinical, imaging, and pathological characteristics, providing valuable practical experience for clinicians and radiologists.

Funding

No

Conflict of Interests

The authors declare that there is no conflict of interest regarding the publication of this paper.

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