Case Report

Multinodular Hydropic Leiomyoma in a 41-Year-Old Patient: A Case Report

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Case Report

Multinodular Hydropic Leiomyoma in a 41-Year-Old Patient: A Case Report

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Abstract

Uterine leiomyomas are a heterogenous group of benign mesenchymal tumors. While diagnosis is usually achieved through clinical assessment and pelvic ultrasound (PU), atypical subtypes are not as easily recognizable and can be mistaken for malignant tumors such as leiomyosarcoma or ovarian carcinoma. We describe the case of a 41-year-old patient who presented with increasing bulk symptoms, urinary frequency and growth of a hydropic leiomyoma of the left lateral and posterior uterine wall that had been known for 10 years, confirmed with previous biopsy. The tumor filled the entire pelvic cavity in PU and was increasingly difficult to delineate. Considering that the patient had no further desire for future pregnancies, an abdominal hysterectomy without oophorectomy was performed. The patient suffered from an iatrogenic injury to the left ureter and underwent ureteroneocystostomy, from which she recovered fully. Gross tissue examination showed an irregularly enlarged, asymmetric uterus with an intrauterine subserosal mass and an extrauterine papillary tumor arising from the right and posterior uterine wall. The tumor measured 20 x 17 x 10 cm in size. Numerous smooth muscle nodules were observed within the uterus and extending into the extrauterine component in a continuous transition, exhibiting a benign, bland appearance. The nodules were separated by abundant edematous connective tissue with increased vascularization. Histopathological analysis revealed low mitotic activity with no evidence of nuclear atypia, pleomorphism, or necrosis. Immunohistochemical staining confirmed the diagnosis of a benign smooth muscle tumor. The findings suggest a rare, benign smooth muscle neoplasm with both intrauterine and extrauterine involvement.

Keywords: hydropic leiomyoma; uterine fibroids; multinodular; perinodular hydropic degeneration; leiomyoma variant; case report

1. Introduction

Uterine leiomyomas, also known as uterine fibroids, are amongst the most common benign gynecologic tumors. Their growth is associated with hormonal factors and their incidence is more common with increasing age. Other risk factors such as family history, environmental factors and race are also thought to play a role [1–3].

It is estimated that only a quarter of leiomyomas lead to symptoms, such as heavy menstrual bleeding, pelvic pain, infertility, urinary symptoms or dyspareunia [4]. The intensity and type of symptoms depend on the size, amount and location of myomas.

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Diagnosis is usually made through physical examination and pelvic ultrasound (PU) alone. Sometimes this basic imaging must be complemented by further examinations, such as sonohysterography, hysteroscopy [5–8] or, in complex cases, by magnetic resonance imaging (MRI). If malignancy is suspected, computer tomography (CT) scans and positron emission tomography (PET) are usually performed to help differentiate between leiomyomas and leiomyosarcomas or ovarian tumors, even though this has not been proven to be sufficiently reliable [9–13]. It is not always possible to obtain histopathological confirmation during the diagnostic stage, which increases the difficulty of decisions regarding therapeutic management.

Therapeutic options range from symptom-oriented and hormonal approaches, uterus-preserving interventions such as myomectomy by laparoscopic or hysteroscopic resection, uterine artery embolization as well as radiofrequency ablation, to radical approaches in the form of hysterectomy [14]. The patient's desire for future pregnancies must be considered when discussing these options.

Due to the advances in the fields of genomics and molecular pathology, an increasing number of histological subtypes have been found in the heterogeneous group of uterine leiomyomas, which is also reflected in the 2014 WHO classification of mesenchymal uterine tumors [15]. In this case report, we present a very rare subtype of uterine leiomyomas, namely multinodular hydropic leiomyomas (HLM), and discuss challenges in imaging, histopathological assessment, management and postoperative outcomes.

2. Case

We present the case of a 41-year-old secundipara who had been diagnosed with a myoma 10 years prior. Initially located in the left lateral uterine wall, measuring around 4 to 5 cm and resembling a typical leiomyoma in PU, the patient was mostly asymptomatic until 6 years after the initial diagnosis had been made. Around this time, she was referred to our hospital group for a second opinion.

The patient, who was 37 years old at this time, complained of urinary frequency, which had been long present, and heavier menstrual bleeding since her first vaginal birth a year prior. Personal and family history was negative for gynecological carcinomas. The remaining past medical and surgical history was unremarkable. PU showed an enlarged uterus with deviation to the right and a homogenous mass of around 10 x 6 cm on the left lateral wall of the uterus from the insertion point of the left uterine artery to the fundus with protrusions into the pouch of Douglas. The ventral, fundal, subserous junction points of the uterus to the mass showed atypical, cystic, papillary structures with increased vascularization (Figure 1). A PET as well as a CT scan of the abdomen and thorax were performed in order to distinguish between a possible leiomyosarcoma and they showed no suspicion of malignancy. At this point in time, the patient was still open to trying for a second pregnancy, so watchful waiting was agreed upon.

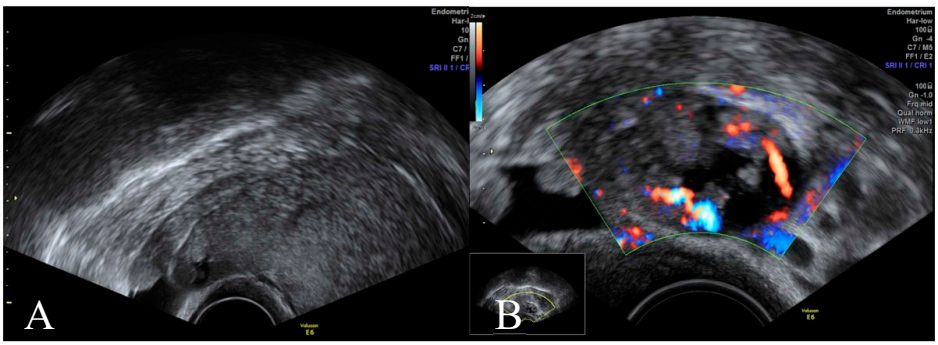

Figure 1. *Pelvic ultrasound (PU) at the first consultation*. Uterine mass suggestive for leiomyoma, reaching from the left lateral wall to the fundus (A). Doppler ultrasound shows a highly vascularized cystic, papillary structure attached to the mass (B).

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At the next consultation a few months later, she had conceived spontaneously. The first and second trimester were free of complications; the leiomyoma remained stable in size. However, at 38 weeks 4 days of gestation, the patient presented at our department with severe itching; cholestasis of pregnancy was diagnosed. The myoma was reevaluated as well due to worsening pelvic pain. Abdominal ultrasound (AU) and PU were suggestive of a slight enlargement of the myoma to 11 x 10 x 10 cm as well as a deviation of the cervix to the right. (Figure 2). We recommended early delivery due to cholestasis of pregnancy via caesarean section (CS) due to obstruction of the labor canal.

CS was performed at 38 weeks 5 days of gestation without any complications. The subserous myoma, measuring around 10 cm, contained attached macroscopic papillary structures in the posterior uterine wall, which were biopsied intraoperatively (Figure 3). Histology confirmed a multinodular HLM.

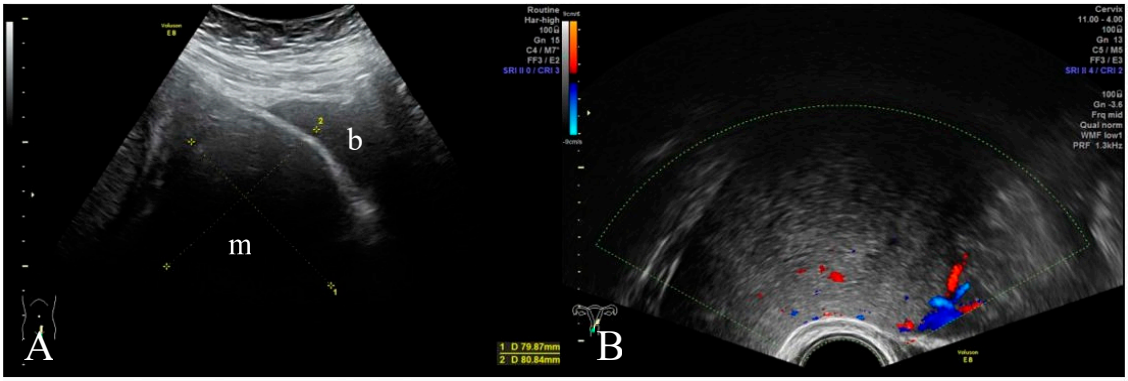

Figure 2. *Uterine mass during pregnancy*. Suspicion of growth of the mass in AU at 38 weeks of gestation (A). Ultrasonographic findings were suggestive of deviation of the labor canal, especially as seen in PU (B). b – bladder, m – mass.

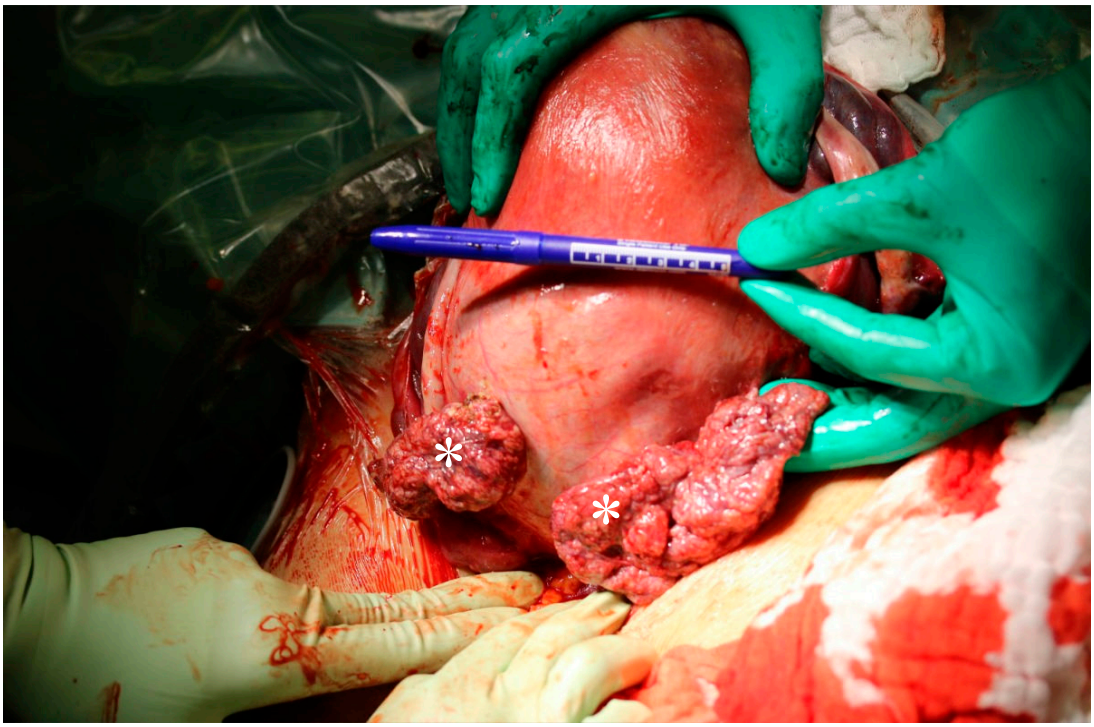

Figure 3. *Intraoperative findings during caesarean section*. The posterior uterine wall shows papillary structures (*) that were attached to the subserous mass. Biopsies were taken and confirmed a benign histology in the form of a hydropic leiomyoma (HLM).

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Over the following two and a half years, the patient presented for several consultations. She initially had a desire for fertility-sparing options with no bulk symptoms, dysmenorrhea or hypermenorrhea. The myoma remained stable in size, around 11 x 7 x 10 cm, and AU showed no suspicion of hydronephrosis. Due to the overall slow progression of the tumor, benign biopsy and oligosymptomatic patient, watchful waiting was agreed upon once again. However, she developed bulk symptoms thereafter, especially involving the bladder, as well as worsened urinary frequency and dysmenorrhea. The myoma had grown to 13 x 13 x 15 cm and PU and AU were increasingly difficult to interpret, as the mass filled the entire pelvis (Figure 4). She was counseled for abdominal hysterectomy, since fertility-sparing options were no longer desired and the growing mass required definitive treatment. 10 years after the initial diagnosis of the myoma and at the age of 41, the patient agreed to undergo abdominal hysterectomy without oophorectomy, and consent was obtained.

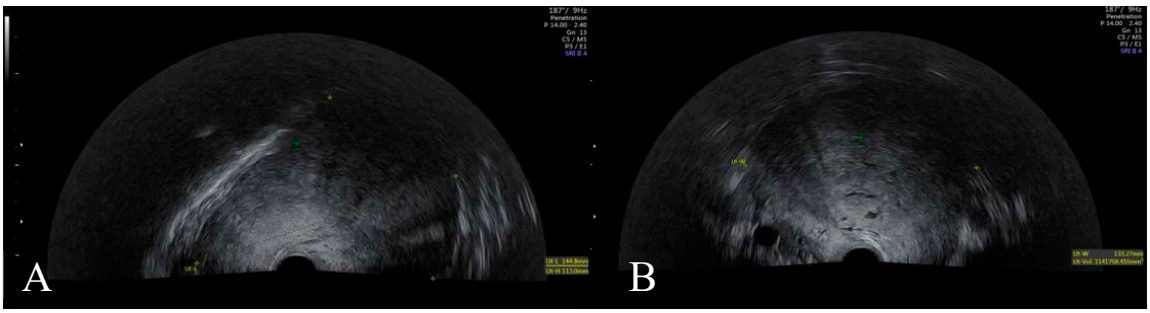
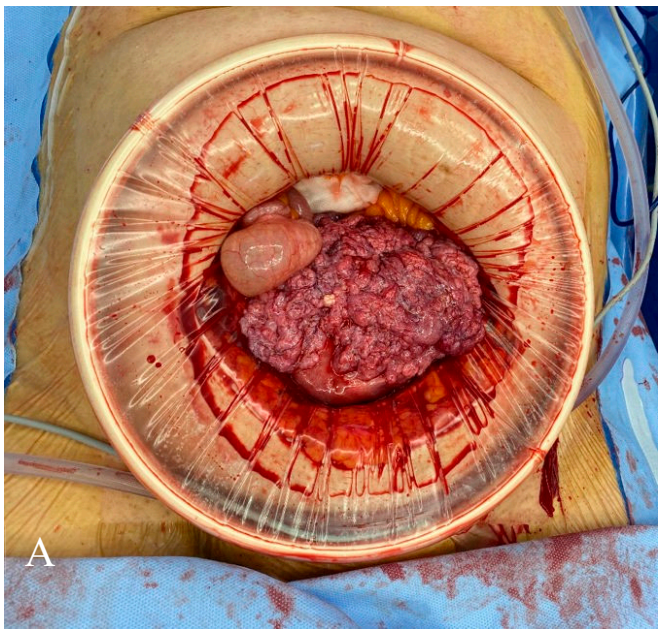


Figure 4. *Growth progression of the leiomyoma*. Two years after the caesarean section, the myoma displayed signs of growth progression and was increasingly difficult to delineate in sagittal (A) and transversal view of PU (B).

Macroscopic examination of the uterus was unusual upon entering the peritoneal cavity, with cauliflower-like tissue protruding from the posterior uterine wall. Despite the past benign histology, a leiomyosarcoma could not be clinically ruled out (Figure 5), although on inspection, the peritoneal lining as well as all other intra-abdominal organs appeared normal and free of carcinomatosis. Several myomas in the left parametrium enlarged the uterus. The hysterectomy was complicated due to the inability to fully mobilize the uterus on the left side and therefore increased difficulty in incising the cervicovaginal junction due to the myomas. Ultimately, this was achieved by opening the vagina from the right side and incising towards the left until the uterine artery was clamped and cut through the vaginal opening. Cumulative intraoperative blood loss totaled 900 milliliters.



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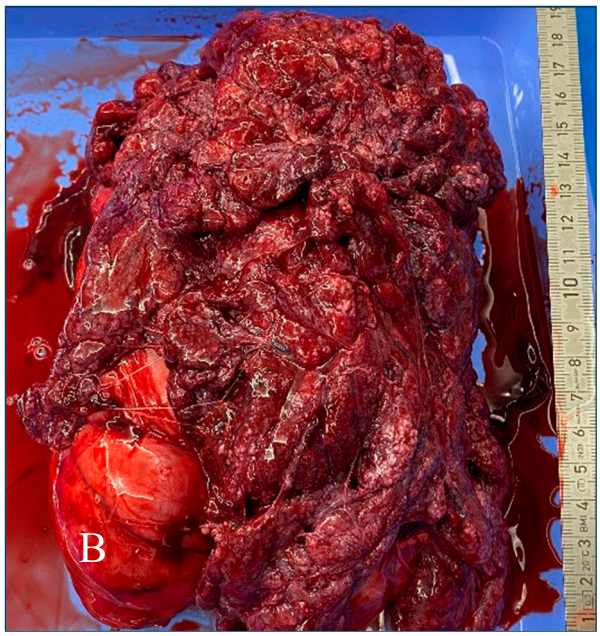

Figure 5. *Intraoperative findings and macroscopy of the uterus.* Macroscopic appearance of the uterus in situ (A) and after hysterectomy before fixation from a frontal (B) perspective.

The patient recovered well despite increased pain during the first night, which required the administration of intravenous opiates. The hemoglobin value was at 110 g/L on the second postoperative day and AU was performed to rule out hydronephrosis before discharge.

Two days after being discharged, she was admitted to the emergency room with strong flank pain on the left side. Clinical examination was not suggestive of pyelonephritis, but the estimated glomerular filtration rate (eGFR) was reduced with a rate of 53 ml/min. AU was performed and showed an enlarged left kidney as well as perirenal free fluid. The CT scan confirmed a rupture of the left renal pelvis as well as an abrupt distal ending of the left ureter with no clear cause of obstruction (Figure 6). The patient underwent emergency pigtail catheterization of the left ureter and retrograde pyelography which showed an injury to the distal ureter. Ureteroneocystostomy in Politano-Leadbetter and Psoas-Hitch technique was performed, from which the patient recovered quickly. eGFR returned to > 90 ml/min before the patient was discharged postoperatively after five days.

The patient presented to the gynecologic follow-up consultation almost two months later with complaints of urinary urgency, but no other symptoms. The pigtail catheter had already been removed. Clinical examination showed fully healed colpotomy and abdominal scars. The AU und PU of the kidneys, bladder and pelvis were normal. The patient was referred to the urologists for ongoing care regarding urinary urgency as well as bladder pain, which we suspected was due to the psoas hitch.

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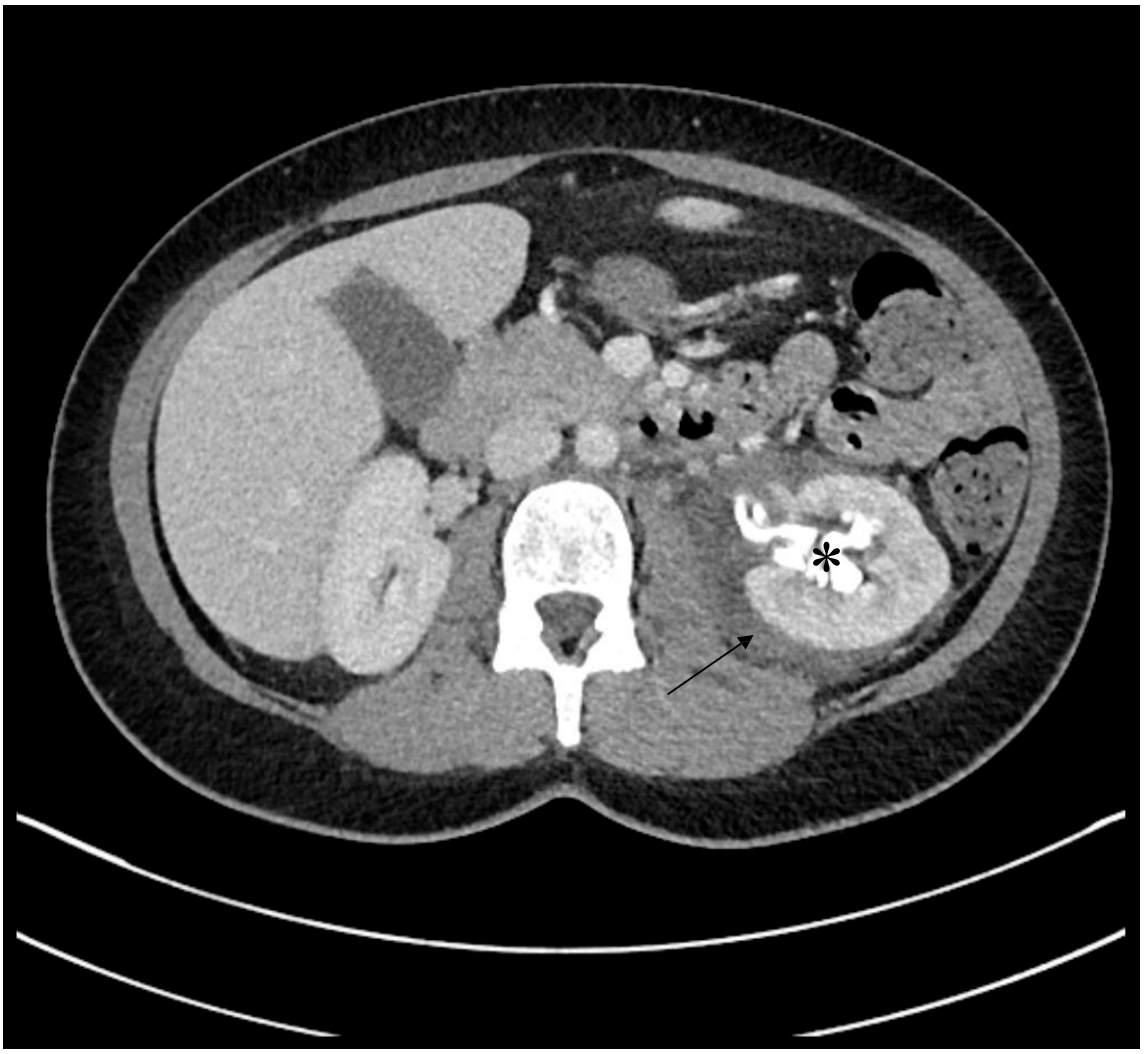

Figure 6. *Postoperative computer tomography (CT) scan.* Perirenal free fluid (arrow) as well as a leakage of contrast agent is seen exiting the left renal pelvis (*), confirming pyelon rupture of the left kidney.

3. Histopathology

The pathological tissue examination of the hysterectomy specimen revealed a multinodular deformation in the cervix and corpus area, with a maximum diameter of 10 cm. Additionally, an exophytically growing, papillary tumor with a brown to livid color was observed at the lateral and posterior corpus site, which contained several solid, white nodular components (Figure 7). The cut surfaces of both the intrauterine and extrauterine tumor areas exhibited a similar appearance, characterized by a white to brown coloration with a cribriform pattern, organized into numerous nodules of varying sizes and showing discharge of serous fluid. The tumor in the myometrial tissue of the uterus was poorly demarcated. Furthermore, small focal adenomyosis was observed, without any association with the smooth muscle neoplasia. Additionally, two normally structured fallopian tubes were identified, each with attached paratubal cysts, the largest of which measured up to 2.5 cm in diameter.

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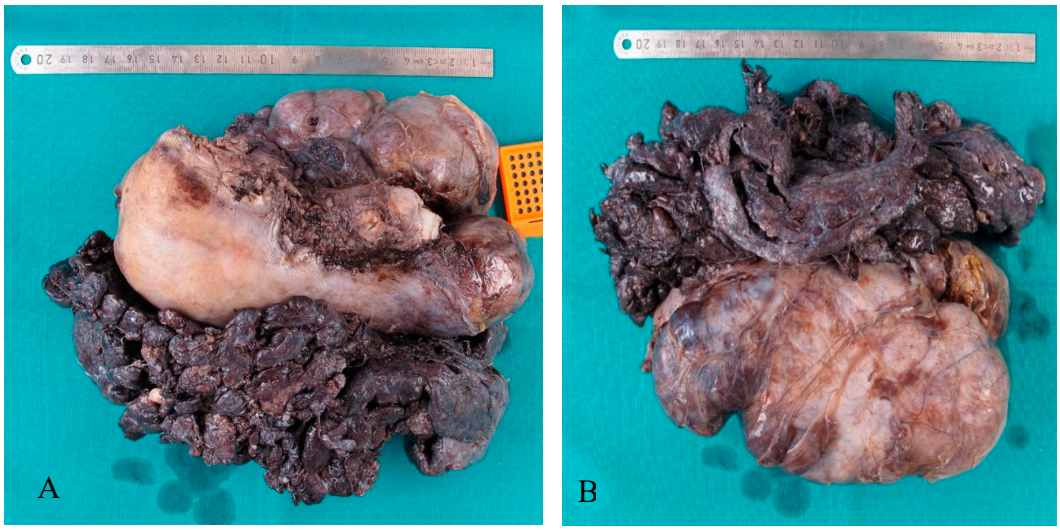

Figure 7. *Hysterectomy specimen after fixation.* Macroscopic appearance of multinodular HLM, displaying the exophytic, papillary fraction on the right and posterior uterine wall, as well as the nodular fraction on the posterior and left uterine wall. (A) Ventral view. (B) Dorsal view.

The neoplastic lesion predominantly consisted of spindle-shaped cells with round to oval nuclei arranged in nodular fascicles, exhibiting a swirled, disorganized pattern (Figure 8). These nodules were separated by abundant edematous, hypocellular stroma (perinodular hydropic degeneration) [16] and displayed an infiltrative growth pattern, with the tumor extending into the surrounding myometrium and causing tissue dissection (Figure 9). Some nodules in the exophytic tumor compound, corresponding to the macroscopically more solid, whitish parts, showed increased hyalinization (Figure 10). A subset of the nodules exhibited perinodular retraction of the hydropic fibroconnective tissue, which may have resembled intravascular infiltration (Figure 11). Immunohistochemical analysis revealed that the neoplastic cells were positive for smooth muscle antigen, desmin, and caldesmon (Figure 12). CD34 showed no endothelial cells delineating individual tumor nodules. No lymphangiosis was observed with D2-40. Tumor cells were negative for CD10.

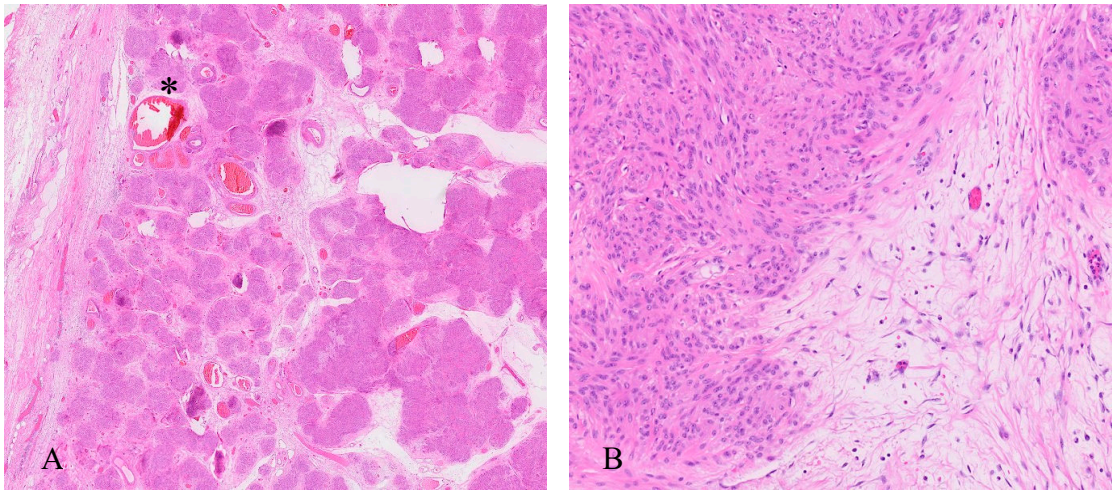

Figure 8. *Histopathology*. (A) Subserosal leiomyoma localized in the cervix with a multinodular appearance, arranged as fascicles of smooth muscle and separated by abundant watery-edematous stroma and containing congested vessels (*). (B) High magnification microscopic image: The smooth muscle cells are arranged in a swirling pattern. The cells are bland, spindle-shaped to oval, with no nuclear atypia or increased mitotic activity. The connective tissue between the muscle bundles appears hydropic, with few fibroblasts.

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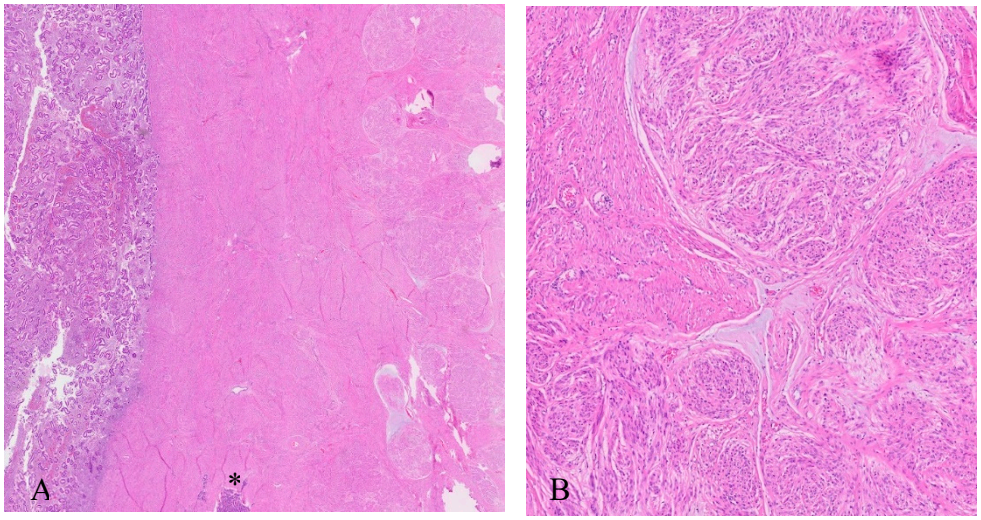

Figure 9. *Histopathology*. (A) HLM dissecting the myometrium of the uterus (right side of image). Intact secretory endometrium (left side). Adenomyosis uteri (*). (B) High magnification showing the HLM dissecting normal myometrial smooth muscle fascicles as well as hydropic changes within the myometrium.

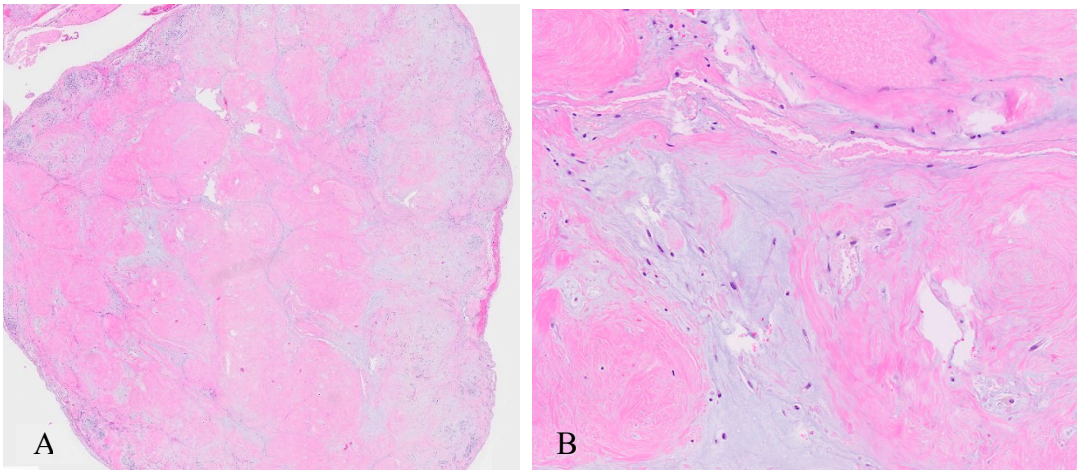

Figure 10. *Histopathology*. (A) Solid portion of the exophytic component of the multinodular HLM with pronounced hyalinization. (B) Higher magnification showing hyalinized smooth muscle fiber bundles separated by hypocellular, watery stroma.

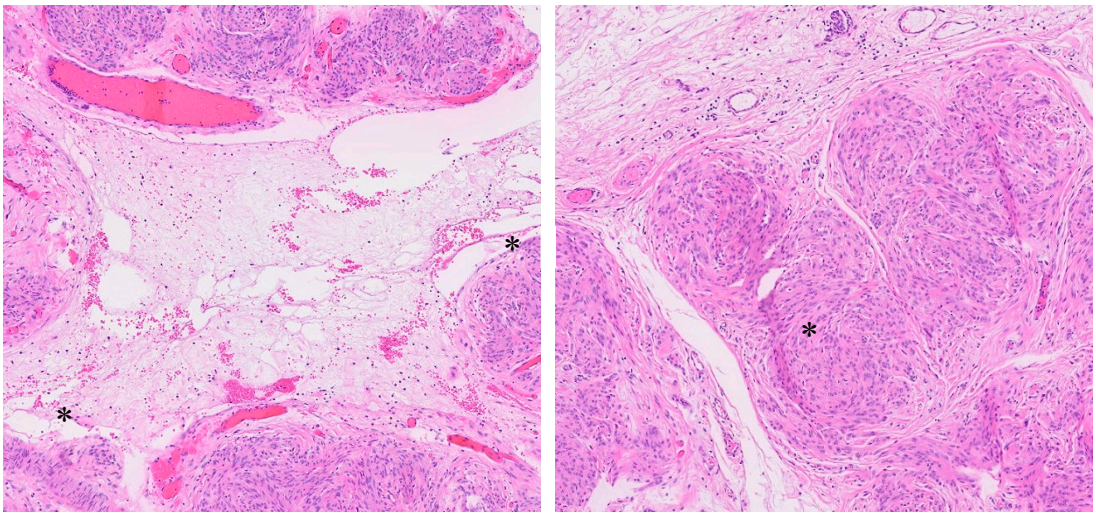

Figure 11. *Histopathology*. Smooth muscle nodules with perinodular hydropic degeneration with artefactual retraction of the intervening stroma (*), creating gaps that may mimic vascular infiltration.

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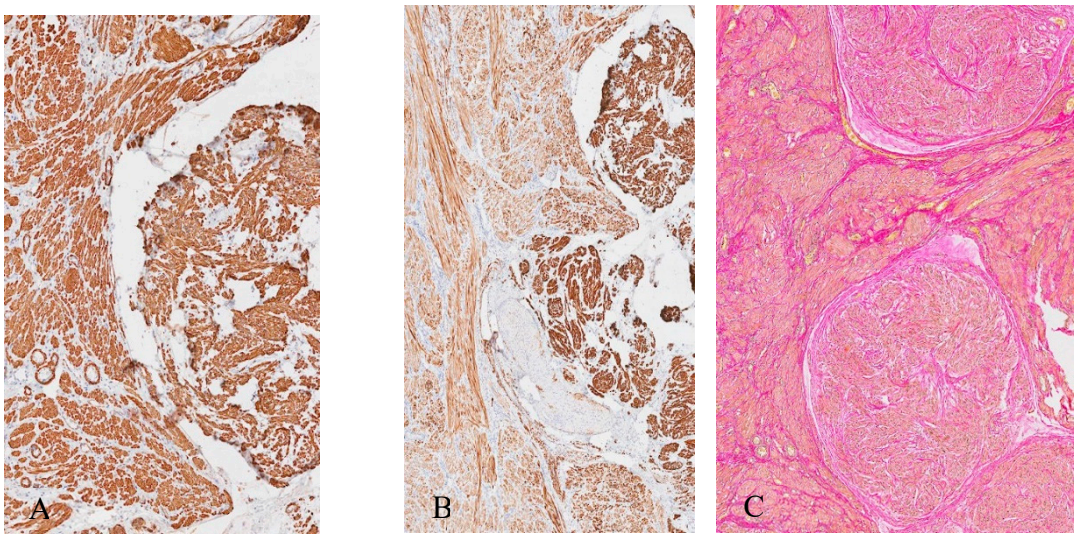

Figure 12. *Immunohistochemical and special stains.* (A) Actin is strongly positive in the myometrium and leiomyoma nodules. (B) Desmin is positive in myometrium and strongly positive in leiomyoma nodules. (C) EvG stain for the visualization of muscle tissue and blood vessels.

4. Discussion

The pathogenesis of HLM is still poorly understood. Advanced analyses like molecular and genetic testing have been described in a few studies, but occurrences remain rare, which hinder new insights [17,18].

Diagnostic imaging of HLM poses a challenge, as it is often inconclusive. Some features of HLM, such as rapid growth and complex cystic structures, may be mistaken for other benign or malignant tumors of the ovary or leiomyosarcoma [16,18,21–23]. Pelvic ultrasound may not reveal characteristic features such as watery edema [19,20], while MRI provides better tissue contrast but cannot definitively exclude malignancy before surgical intervention [20,24,25]. Although diffusion-weighted imaging and the "split fiber" sign have shown promise [26], securing histopathology in a timely manner remains the safest and most reliable approach [19,21].

This hydropic multinodular leiomyoma exhibits distinct perinodular hydropic degeneration, which divides the tumor into numerous smooth muscle nodules separated by watery, edematous connective tissue, creating its characteristic multinodular pattern. This degeneration can resemble intravascular infiltration or intravascular leiomyomatosis, especially when there is severe degeneration with retraction [16,27–31], which can be ruled out by immunohistochemical staining of endothelial cells (CD34).

Another set of differential diagnoses for HLM are low-grade endometrial stromal sarcoma and myxoid leiomyosarcoma. The absence of CD10 expression and lymphovascular invasion excluded the former, while the absence of atypia, low mitotic activity (1%), lack of nuclear atypia, pleomorphism, necrosis and myxoid stroma ruled out the latter [29,32–34]. Other rare variants, such as cotyledonoid or intravenous leiomyomatosis, may show overlapping features [16,29–31], with some authors regarding them as different phases of evolutionary changes.

As a single case report, this study has limited impact on a wider scale and our findings as well as our therapeutic management are not necessarily generalizable; the rare occurrence of this disease contributes to this fact. However, individual contributions to the literature regarding atypical leiomyoma presentation and histology are helpful to fill gaps and encourage further studies as well as raise awareness for these possible differential diagnoses, so that the best diagnostic and therapeutic decisions can be made available to patients.

In our case, the gradual growth, inconclusive imaging, and extrauterine expansion led to surgical intervention despite prior benign histology, which is not always available in similar cases.

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Our experience highlights the need for individualized management of atypical leiomyomas as well as the importance of expert surgical planning in complex cases, particularly when diagnostic or surveillance imaging is inconclusive.

5. Conclusions

Uterine mesenchymal tumors represent a heterogeneous group. In challenging cases with marked tumor growth and inconclusive imaging, surgery remains the first-line treatment. While hydropic leiomyomas are benign entities, they can closely mimic malignant tumors, which can only be ruled out through thorough histopathological evaluation. Careful pre- and intraoperative management is essential for optimal treatment of such rare variants.

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