



A Rare Large Cystic Adenomatoid Tumor of the Uterus: Diagnostic Challenges and Focused Literature Review

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Abstract

Uterine Adenomatoid Tumor (AT) is a rare benign mesothelial neoplasm, usually small and incidental. Large cystic presentations are exceptional and can mimic adnexal or uterine malignancy. The Leiomyoadenomatoid Tumor (LAT) variant combines mesothelial and smooth-muscle components, adding diagnostic difficulty. The Leiomyoadenomatoid Tumor (LAT) is a rare benign uterine lesion characterized by a proliferation of interlacing smooth muscle fibers admixed with glandular elements and fibrous tissue, forming well-demarcated nodules. A 53-year-old postmenopausal woman presented with diffuse pelvic pain and one month of progressive abdominal distension. Preoperative laboratory tests, including measurement of serum CA-125, were within normal limits. No elevation of tumor markers was observed, supporting the benign nature of the lesion and helping to exclude malignant ovarian or uterine tumors in the differential diagnosis. Ultrasound revealed a 16 × 10 cm unilocular anechoic cystic lesion, and pelvic MRI showed a large heterogeneous solid-cystic uterine mass with low T1/T2 signal in the solid part, high T1 hemorrhagic areas in the cystic part, and moderate peritoneal effusion, contiguous with the posterior uterine wall. Serum CA-125 was within normal limits. Laparotomy identified a 17-cm cystic mass attached to the right uterine wall, and total hysterectomy with bilateral adnexectomy was performed. Histopathology demonstrated intersecting bundles of bland spindle cells (smooth muscle) with a peripheral cyst lined by attenuated mesothelial epithelium, without atypia, necrosis, or significant mitotic activity (1/10 HPF). Cytology of hemorrhagic peritoneal fluid showed benign mesothelial clusters. Immunohistochemistry (cytoblock) was calretinin-positive and PAX8-negative, confirming mesothelial origin. TRAF7 mutational testing was not performed in this patient, which represents a limitation

of this case report. **Postoperative Course and Long-Term Follow-Up:** The postoperative course was uneventful, with no hemorrhagic, infectious, or thromboembolic complications. Pain was well controlled, and recovery was rapid. Clinical and pelvic ultrasound follow-up at three and six months showed no recurrence or local complications. The patient remained asymptomatic, and the CA-125 level was within the normal range, confirming the favorable prognosis and benign behavior of the Leiomyoadenomatoid Tumor. LAT is a rare benign uterine lesion that may present as a large symptomatic mass mimicking malignancy. Recognition of its dual mesothelial and smooth-muscle nature through histopathology and immunohistochemistry is essential to avoid overtreatment. Surgical excision is curative, and prognosis is excellent. This study was conducted in accordance with the ethical principles of the Declaration of Helsinki. Institutional ethics committee approval was obtained, and written informed consent was obtained from the patient for publication of the clinical data and related images.

Subject Areas

Oncology

Keywords

Leiomyoadenomatoid Tumor, Uterus, Mesothelial Neoplasm, Cystic Pelvic Mass, MRI, Immunohistochemistry, TRAF7, Case Report

1. Introduction

A adenomatoid tumor is an uncommon benign mesothelial neoplasm of the genital tract. In women, it typically arises from the subserosal myometrium or serosal surfaces (peritoneum/mesosalpinx) and is most often an incidental small nodule discovered during surgery for other indications [1]. Histologically, adenomatoid tumors show tubular/microcystic mesothelial proliferations with bland cytology and low mitotic activity and express calretinin, WT1, CK5/6, and D2-40, while PAX8 is negative—helpful to exclude Müllerian lesions [2]. Although the typical lesion is small, atypically large or cystic uterine presentations are rare and diagnostically challenging, with MRI features recently described in dedicated series [3]-[7] and case compilations focusing on the cystic variant [8]. On the molecular side, TRAF7 mutations have been identified across genital-tract adenomatoid tumors, supporting a clonal benign neoplasm rather than a hamartoma and informing difficult differentials [9]-[11]. Misdiagnosis remains common, particularly as a degenerating leiomyoma or adnexal tumor [12].

2. Case Presentation

A 53-year-old G2P2 (two prior cesarean deliveries), menopausal for 12 years, presented with diffuse pelvic pain and rapidly progressive abdominal distension over

one month. General examination was unremarkable; the abdomen was moderately distended with a centrally located, slightly tender pelvic mass. The Pfannenstiel incision scar appeared well healed.

Pelvic ultrasound showed a well-defined unilocular anechoic cystic lesion measuring 16×10 cm without septa or mural nodules. Pelvic MRI demonstrated a mixed solid-cystic mass arising from the posterior uterine body, extending cranially toward the right hypochondrium (maximal dimensions $216 \times 154 \times 94$ mm), displacing adjacent structures without invasion (**Figure 1**). Differential diagnoses included a large exophytic degenerating leiomyoma versus an adnexal mass of uncertain origin.

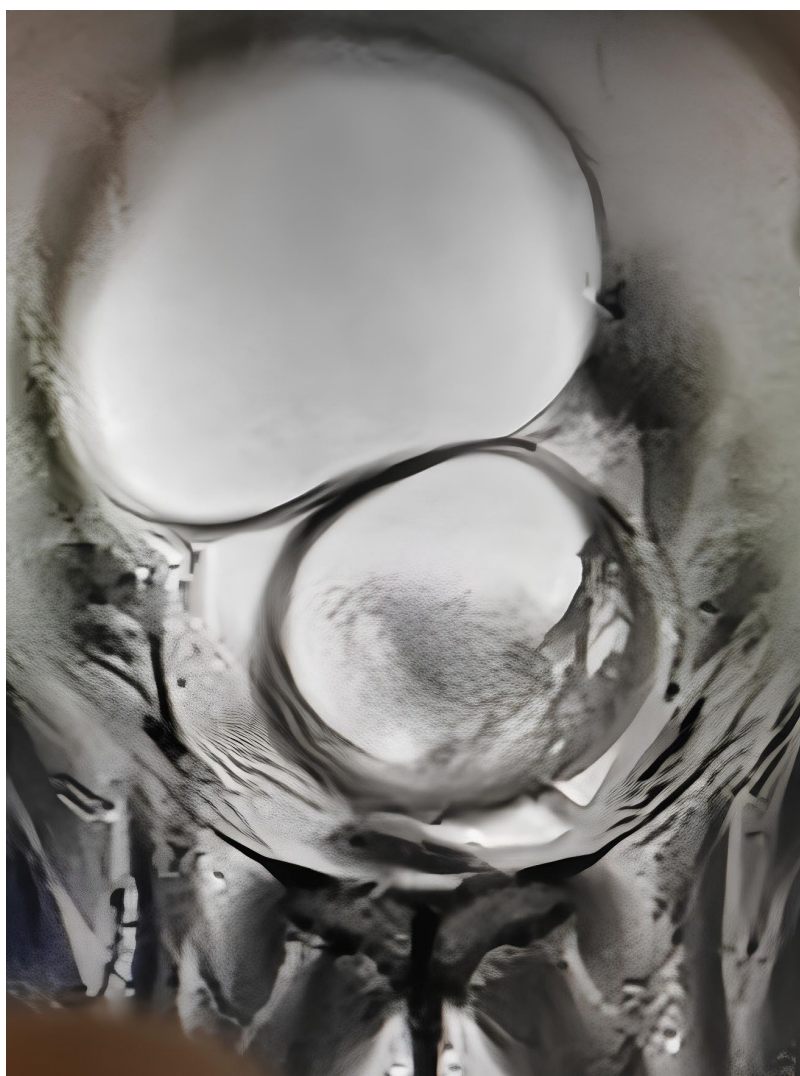


Figure 1. Pelvic MRI showing a large mixed solid-cystic mass contiguous with the posterior uterine wall (max. $216 \times 154 \times 94$ mm), displacing adjacent viscera without invasion.

Surgery: exploratory laparotomy identified a 17-cm cystic mass attached to the right uterine wall, with a highly vascular external surface and hemorrhagic intracystic fluid, without adhesions or organ invasion (**Figure 2**).



Figure 2. Intraoperative view of a 17 cm cystic mass attached to the right uterine wall; vascularized outer surface. The black arrow indicates the inclusion cyst, and the red arrow indicates the uterus.

Given the patient's postmenopausal status, tumor size, and uncertain preoperative diagnosis, total hysterectomy with bilateral adnexectomy was performed. Peritoneal fluid was sampled for cytology (**Figure 3**). Postoperative recovery was uneventful.

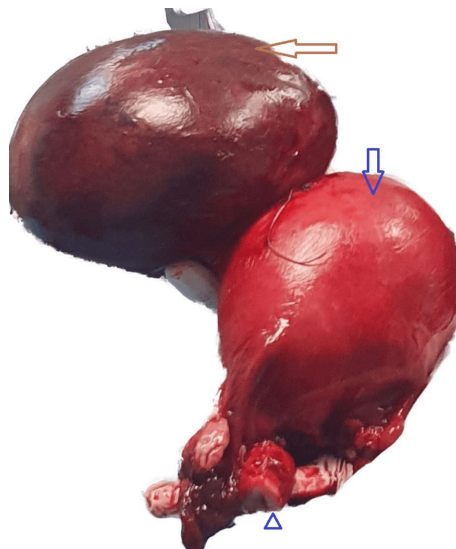


Figure 3. Gross specimen: uterus with attached cystic mass; cut surface revealing hemorrhagic cyst contents. The brown arrow shows the cyst, the indigo arrow shows the uterus, and the indigo triangle indicates the cervix.

The hysterectomy specimen showed a benign spindle-cell proliferation arranged in short intersecting bundles (smooth muscle), no atypia, and 1 mitosis/10 HPF, without necrosis or intratumoral hemorrhage. The endometrium was atrophic; cervix and adnexa were unremarkable (**Figure 4**).

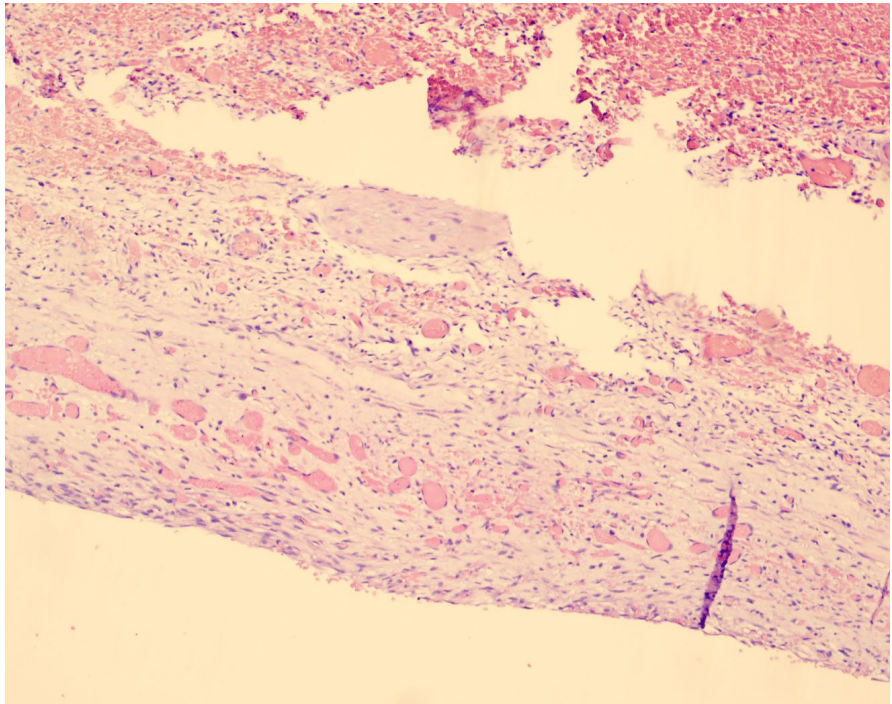


Figure 4. Microscopic image of the cyst. Inclusion cyst ($\times 10$, coloration with hematoxylin and eosin).

At the periphery of the mass, there was a well-circumscribed cyst with a fibrous wall lined by attenuated mesothelial epithelium, largely denuded, and without communication with the smooth-muscle bundles (**Figure 5**).

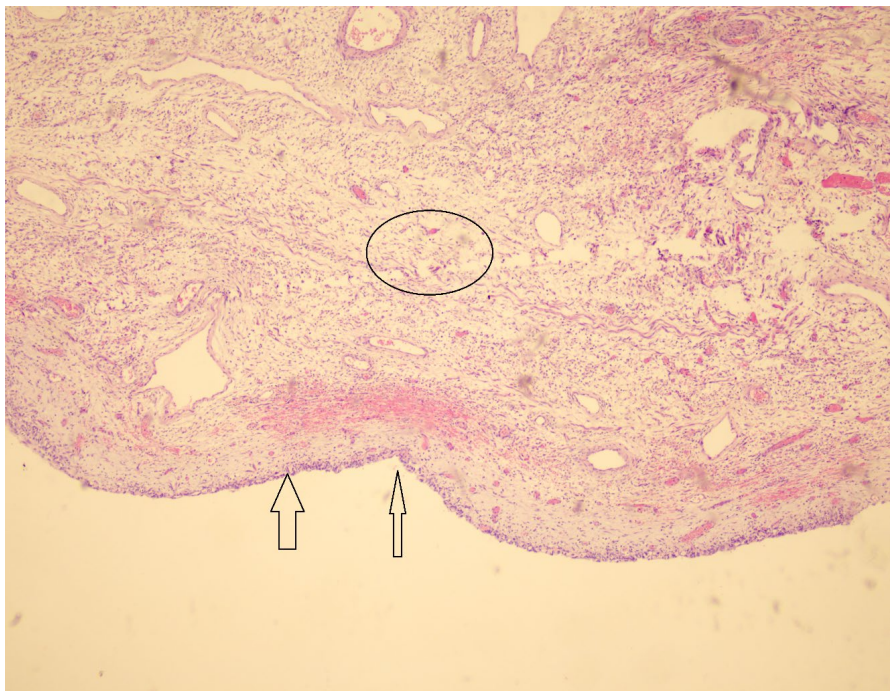


Figure 5. Inclusion cyst: fibrous wall (outlined with black oval) lined by mesothelial cells (two black arrows). $\times 4$, coloration with hematoxylin and eosin.

These features support a benign mesothelial lesion with prominent smooth-muscle stroma. Peritoneal fluid cytology showed a hemorrhagic background with small pseudoglandular clusters of mesothelial cells with minimal atypia and no malignant features (**Figure 6** & **Figure 7**).

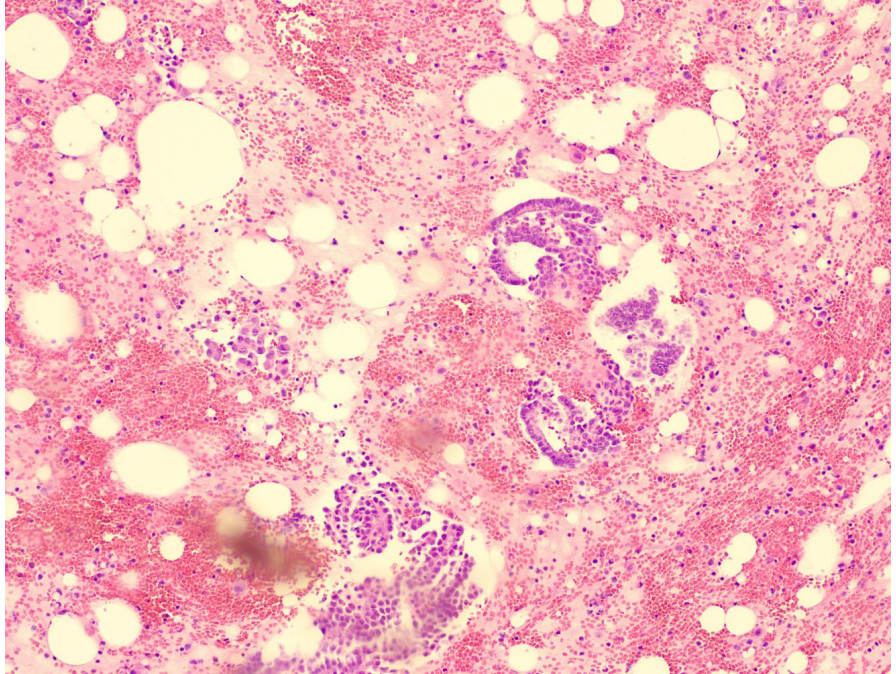


Figure 6. Microscopic appearance of the ascitic fluid. Pseudoglandular clusters of mesothelial cells in a hemorrhagic background. $\times 4$, coloration with hematoxylin and eosin.

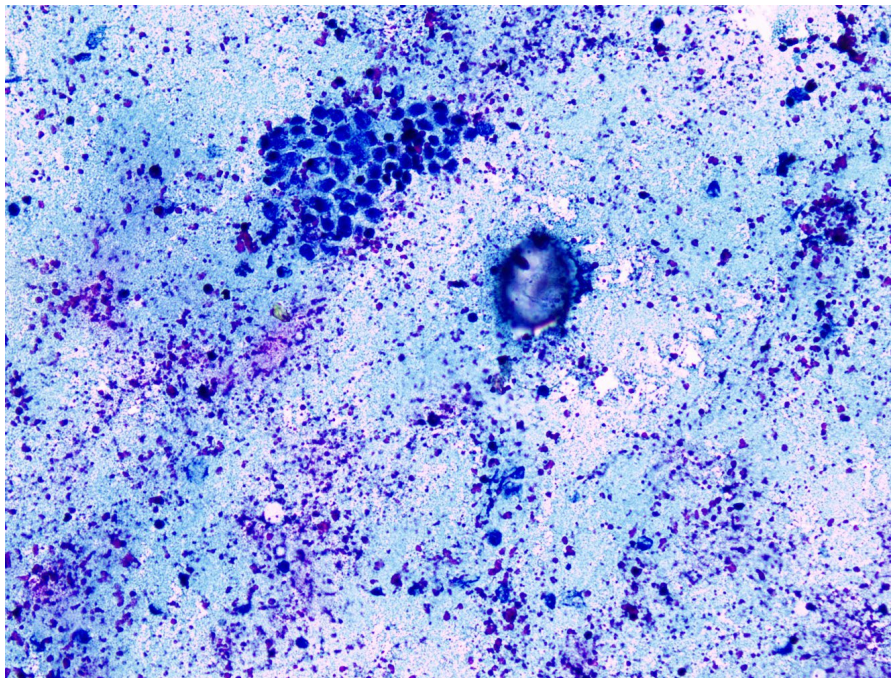


Figure 7. Clusters of mesothelial cells in the ascitic fluid stained with Papanicolaou. Magnification $\times 20$.

Immunohistochemistry performed on a cytoblock was calretinin-positive and PAX8-negative, corroborating mesothelial lineage and excluding Müllerian epithelium (**Figure 8** & **Figure 9**).

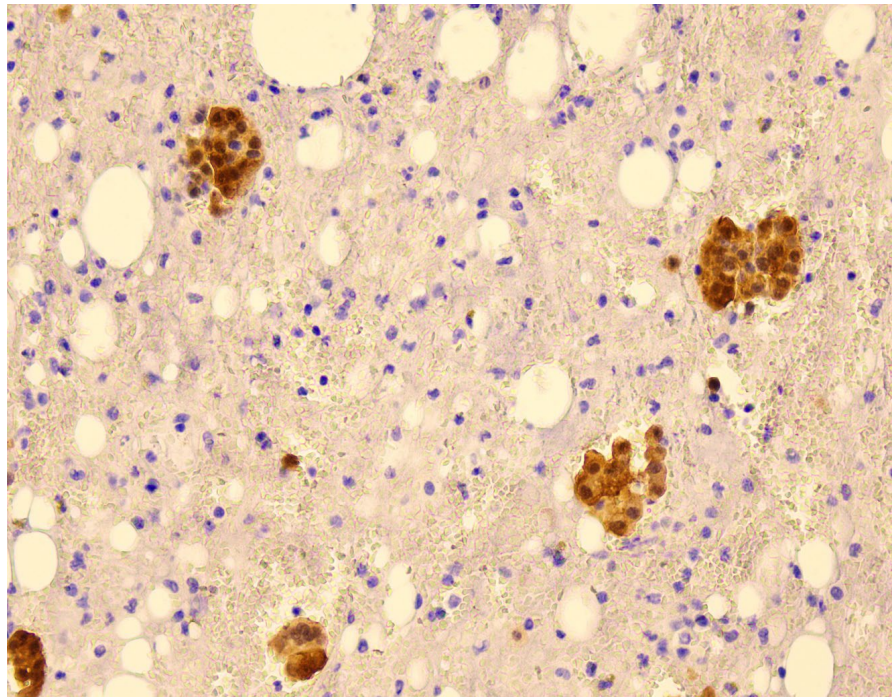


Figure 8. Immunohistochemical analysis showing immunoreactivity of mesothelial cells with anti-calretinin antibody. Magnification $\times 20$.

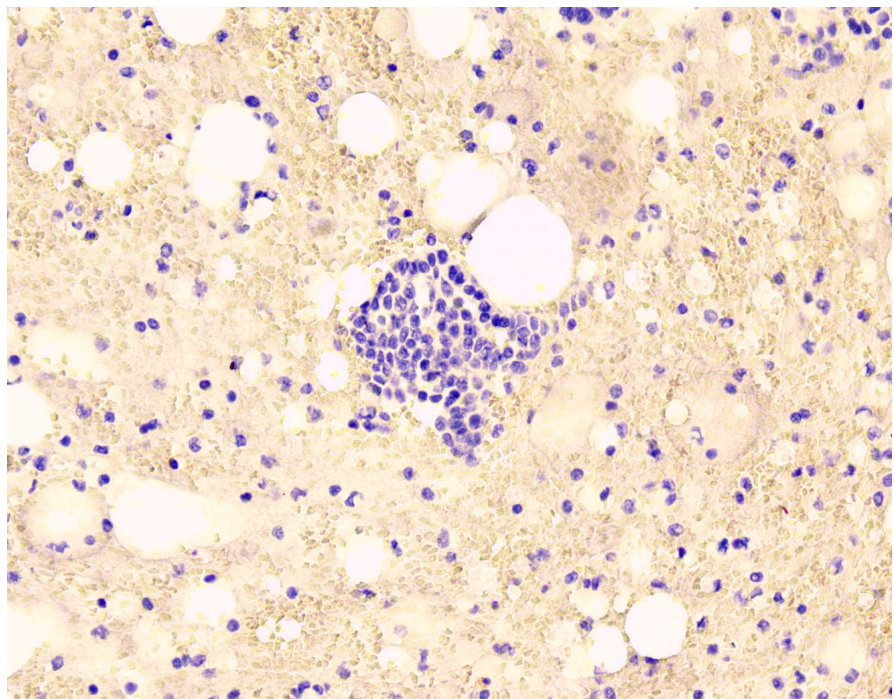


Figure 9. Immunohistochemical analysis showing no immunoreactivity of mesothelial cells with anti-PAX8 antibody. Magnification $\times 20$.

3. Discussion

Uterine adenomatoid tumor is usually small, subserosal, and asymptomatic [1]. Large and cystic forms, although uncommon, can blur the distinction from degenerating leiomyoma, ovarian cystadenoma, or even metastatic adenocarcinoma when imaging is non-specific [3]-[8]. Recent MRI series describe heterogeneous T2 signal with mixed low-signal myoma-like areas and higher-signal microcystic components, sometimes with a peripheral rim, features that can raise suspicion for adenomatoid tumor when uterine continuity is evident [6] [7]. Misdiagnosis at presentation remains frequent [12], and giant cystic uterine adenomatoid tumors have been reported, underlining the potential for size-related symptoms and malignant mimicry [13].

3.1. Pathogenesis and Immunophenotype

The characteristic mesothelial immunoprofile (calretinin, WT1, D2-40 positive; PAX8 negative) distinguishes adenomatoid tumor from Müllerian lesions [2]. The discovery of TRAF7 mutations across genital-tract adenomatoid tumors supports a clonal, benign neoplasm with NF- κ B pathway involvement and can aid in difficult differentials (e.g., atypical growth patterns or nerve adjacency) [9]-[11]. Associations with iatrogenic immunosuppression have been reported and may influence detection or growth [14] [15].

Adenomatoid tumors demonstrate diverse clinical presentations and anatomical distributions. While most commonly encountered in the uterus and fallopian tubes [16], these tumors can present with synchronous involvement of multiple sites [17] or multicentric patterns affecting the uterus, ovary, and appendix [18]. Clinical associations with systemic conditions have been documented, including cases occurring in patients with chronic renal failure [19]. Rare extragenital locations have also been reported, with documented cases in the omentum where adenomatoid tumors may coexist with well-differentiated papillary mesothelioma [20], and exceptionally rare presentations arising from the diaphragm [21]. Understanding this anatomical spectrum is essential for accurate diagnosis and appropriate management.

3.2. Variant with Smooth-Muscle Stroma

When a prominent leiomyomatous component coexists with mesothelial microcysts/tubules, the lesion is referred to as a leiomyoadenomatoid tumor [22]. The present case—spindle-cell bundles plus a peripheral mesothelial-lined cyst—fits this spectrum and helps explain preoperative confusion with leiomyoma.

3.3. Management and Prognosis

Complete surgical excision is curative. There is no malignant potential, and recurrence is exceptional [1]. In postmenopausal patients with large or symptomatic lesions—and when malignancy cannot be confidently excluded—hysterectomy is reasonable; in younger patients with smaller, well-characterized lesions, conservative

excision may be considered.

4. Conclusion

Uterine adenomatoid tumor—occasionally with leiomyoadenomatoid features—can present as a large cystic mass, posing diagnostic challenges. Histopathology with targeted IHC is decisive for confirming mesothelial origin and avoiding over-treatment. Prognosis after complete excision is excellent. This study was conducted in accordance with the ethical principles of the Declaration of Helsinki. Institutional ethics committee approval was obtained, and written informed consent was obtained from the patient for publication of the clinical data and related images.

Conflicts of Interest

The authors declare no conflicts of interest.

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