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Case Report

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Unmasking the Hidden Uterine Cavity: Laparoscopic Excision of ACUM in an Adolescent Girl

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HIGHLIGHTS

- Rare Müllerian anomaly causing severe dysmenorrhea.
- Occurs in young, nulliparous female patients.
- Intramyometrial cyst lined with endometrium.
- Diagnosed through imaging and laparoscopy.
- Surgical excision provides complete symptom relief.

Key Words:

ACUM

Dysmenorrhea

Müllerian anomalies

Laparoscopy

Uterine malformation

ABSTRACT

Introduction: Accessory Cavitated Uterine Malformation (ACUM) is a rare congenital Müllerian anomaly characterized by a non-communicating accessory uterine cavity lined with functional endometrial tissue. It is usually located near the uterine cornua and presents in young, nulliparous women with severe dysmenorrhea and chronic pelvic pain. Due to its rarity and overlapping features with other gynecological disorders, ACUM is often misdiagnosed, delaying appropriate management. Case Presentation: An 18-year-old nulliparous female presented with severe, cyclical lower abdominal pain and dysmenorrhea persisting for three years, unresponsive to conventional medical therapy. Pelvic ultrasonography and magnetic resonance imaging revealed a well-defined intramyometrial cystic lesion near the right uterine horn, not communicating with the endometrial cavity, suggestive of ACUM. Laparoscopic evaluation confirmed the diagnosis, and the lesion was excised completely. Postoperative recovery was uneventful, and the patient reported complete resolution of dysmenorrhea during follow-up. Discussion: ACUM results from a developmental anomaly involving the Müllerian ducts, leading to the formation of an accessory uterine cavity with functional endometrium. The condition mimics endometriosis, adenomyosis, or uterine fibroids on imaging. MRI serves as the diagnostic modality of choice due to its superior soft-tissue characterization. Surgical excision of the accessory cavity is curative and provides long-term symptom relief, preserving fertility. Conclusion: ACUM, though rare, should be considered in young women presenting with refractory dysmenorrhea or chronic pelvic pain. Early recognition and laparoscopic excision result in excellent symptom resolution and improved quality of life.



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INTRODUCTION

Accessory Cavitated Uterine Malformation (ACUM) is a rare congenital anomaly characterized by a cystic lesion within the uterus, lined by functional endometrial tissue and surrounded by hypertrophic myometrium. These lesions typically arise on the lateral aspect of the myometrium, just beneath the insertion point of the round ligament. ACUM predominantly affects young, nulliparous women under the age of 30 and is seldom seen in multiparous individuals [1,2].

Clinical presentation often begins around the time of menarche, with hallmark symptoms including severe dysmenorrhea unresponsive to conventional medical therapy, chronic pelvic pain, and occasionally dyspareunia. The pain tends to exacerbate with the onset of menstruation; however, some patients may remain asymptomatic [3].

The etiology of ACUM is believed to be related to Müllerian duct malformations, potentially linked to gubernaculum dysfunction during embryologic development [4]. Despite increasing recognition in recent literature, the overall understanding of ACUM remains limited, and no large-scale epidemiological studies have been conducted to determine its true prevalence. Consequently, ACUM is frequently underdiagnosed or misidentified as other gynecologic conditions, such as degenerating fibroids, cystic adenomyosis, or congenital uterine anomalies like a bicornuate uterus [5].

In this case report, we describe the presentation, diagnosis, and surgical management of an 18-year-old female diagnosed with ACUM in 2024. This case underscores the need for heightened clinical awareness among gynecologists and provides insight into the optimal management strategies for this uncommon uterine malformation.

CASE PRESEPENTATION

Patient History

An 18-year-old nulliparous woman presented with a three-year history of progressively worsening dysmenorrhea, beginning approximately three years after menarche at age 12. The pain was unrelieved by NSAIDs and often persisted for up to two weeks post-menses. There were no significant medical or surgical antecedents.

Examination and Imaging

Physical and abdominal examinations were unremarkable. Pelvic ultrasound revealed a 24x19 mm hypoechoic lesion with central cavitation and mild internal fluid content, located on the left side of the uterus, receiving blood supply from the uterine artery (Figure 1a). MRI confirmed a T2-hypointense, non-communicating intramyometrial lesion measuring 1.53x1.54x 1.6 cm, suggestive of ACUM (Figure 1b).

Surgical Management

The patient underwent laparoscopic exploration. A spherical lesion was visualized beneath the left uterine horn. After careful dissection, the mass was excised intact (Figure 2). The lesion was filled with chocolate-colored fluid and had a smooth muscular wall lined by endometrial tissue (Figure 3).

Histopathology confirmed the diagnosis of ACUM, revealing endometrial glands and stroma surrounded by smooth muscle fibers without evidence of adenomyosis (Figure 4).

Postoperative Outcome

The patient had an uneventful recovery and was discharged on postoperative day 3. At a three-month follow-up, she reported complete resolution of dysmenorrhea.





Figure 1: (a) USG and (b) MRI showing well circumscribed heterogeneously anechoic area surrounded by thick rim in left lateral aspect

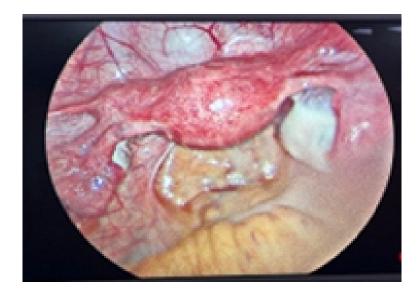


Figure 2: Intraoperative image showing mass beneath left round ligament in laparoscopic view



Figure 3: Gross cut specimen showing uterine cavity

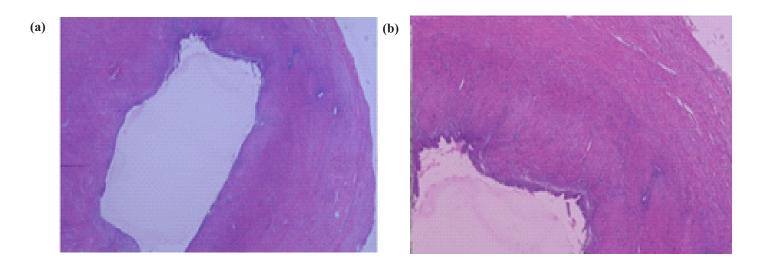


Figure 4: (a, b) Histopathogical examination showed uterine wall with endometrial cavity

DISCUSSION

Previously referred to as "accessory cavitated uterine mass or juvenile cystic adenomyoma", Accessory Cavitated Uterine Malformation (ACUM) is characterized by isolated, cystic lesions in the myometrium. These lesions do not communicate with the uterine cavity and are most commonly observed in nulliparous women under 30 years of age. ACUM typically measures ≥10 mm and is filled with menstrual fluid and blood. The cyst wall consists of uterine smooth muscle and is lined by functional endometrial glands and stroma [6]. Recent studies have demonstrated that ACUM can also occur in adult women, which suggests that the term "juvenile" in its previous nomenclature may lead to confusion [7].

Although both ACUM and juvenile cystic adenomyomas are myometrial pathologies, they share the feature of containing endometrial glands and stroma within the myometrium. However, adenomyosis exhibits a highly variable morphological appearance, while ACUM consistently presents with cysts lined by endometrial glands and stroma. Consequently, the term "malformation" is more appropriate than "mass" to describe the pathogenesis of this condition, as it reflects a developmental anomaly [8].

Recent research has supported the hypothesis that ACUM results from a congenital anomaly of the Müllerian duct. In 2021, the term "accessory cavitated uterine malformation" was proposed to more accurately describe cystic lesions located within the myometrium. These lesions have distinct locations, characteristic symptoms, and specific imaging features.

Diagnostic Criteria

The diagnostic criteria for ACUM, as proposed by Acién et al., are widely accepted and include the following:

- 1. An isolated accessory cavitated mass within the myometrium.
- 2. A normal uterus (with a cavity lined by functional endometrium), fallopian tubes, and ovaries.
- 3. Pathological confirmation post-surgical excision.
- 4. The cystic cavity is filled with chocolate-colored fluid.
- 5. Absence of true adenomyosis.
- 6. The accessory cavity is lined with functional endometrial glands and stroma.
- 7. A concentric arrangement of smooth muscle in the myometrial mantle.

Clinical Presentation

ACUM is most commonly associated with severe dysmenorrhea, which does not respond to conventional pain relief, and recurrent pelvic pain. These symptoms, however, are nonspecific and commonly seen in many gynecological conditions, leading to delayed or incorrect diagnoses. In some cases, ACUM lesions are misidentified as uterine fibroids. For example, in our study of three cases, ACUM was initially misdiagnosed as uterine fibroids. The majority of ACUM lesions are ≤50 mm in size, making it difficult to distinguish from uterine fibroids, fibroid cystic changes, or adenomyosis in ultrasound imaging. In our cases, the average external diameter was found to be 22.8 mm, with a mean internal diameter of 14.1 mm. The small size of the lesions further complicates the diagnosis. The time between the onset of dysmenorrhea and surgery in our cases was three years, which we attribute to the limited awareness of ACUM among both patients and gynecologists [9].

Diagnostic Imaging

Ultrasound, particularly transvaginal ultrasound, is considered the first-line diagnostic tool for ACUM. This approach is favored for its convenience, accuracy, non-invasive nature, and cost-effectiveness. When fully utilized, 3D transvaginal ultrasound can clearly visualize an intramyometrial, rounded cystic structure with sharp edges and finely echogenic fluid content. For nulliparous women, transrectal ultrasound may also be considered [10].

Magnetic Resonance Imaging (MRI) is another effective diagnostic method for ACUM, offering detailed images of intramyometrial hemorrhagic cysts. MRI also provides a precise relationship between the cyst and the surrounding myometrium and/or endometrium without the need for contrast injection. Hypersignal on T1- and T2-weighted sequences indicates hemorrhagic content, while the typical hypointensity of the surrounding tissue on T2-weighted images corresponds to fibrous tissue.

Association with Infertility

While there have been reports of ACUM diagnoses in women with infertility, there is no definitive evidence linking ACUM to reproductive health issues. Most studies on ACUM have been case series, and there is a notable lack of population-based, longitudinal research. The long-term outcomes of ACUM, including its potential impact on fertility and recurrence after surgery, remain unclear.

Management and Treatment

Management strategies for ACUM vary based on symptom severity and patient needs. Conservative treatment options include expectant management, non-steroidal anti-inflammatory drugs (NSAIDs), oral contraceptive pills (OCPs), levonorgestrel intrauterine system (LNG-IUS), and GnRH agonist therapy. Although hormonal treatments may provide temporary relief from dysmenorrhea, symptoms often recur once the therapy is discontinued. Due to the limited efficacy of conservative treatments, surgical intervention is considered the most effective long-term solution, especially in women with severe symptoms.

Fertility preservation is a key consideration in young women, and minimally invasive laparoscopic surgery is the preferred approach. The majority of ACUM lesions are located on the lateral aspect of the myometrium, beneath the round ligament insertion. For this reason, an anterior uterine wall incision is recommended to reduce the risk of injury to the ascending branch of the uterine artery. The boundary between ACUM lesions and the surrounding myometrium is typically well-demarcated, which allows for resections similar to laparoscopic myomectomy [8-10].

Surgical Considerations

Before performing surgery, it is essential to rule out any associated urinary tract anomalies, as these may be present in conjunction with Müllerian anomalies. This step helps reduce the risk of urological injury during surgery.

CONCLUSION

ACUM is a notable cause of dysmenorrhea in young women, though it may often be overlooked or misdiagnosed due to its rare occurrence, non-specific symptoms, and small lesion size. This condition should be considered in patients experiencing severe dysmenorrhea that does not respond to conventional treatments. Timely diagnosis and appropriate management, particularly through laparoscopic minimally invasive surgery, can significantly enhance the patient's quality of life. ACUM is a distinct uterine malformation marked by cystic lesions containing functional endometrial glands and stroma within the myometrium. The clinical presentation, which typically includes severe dysmenorrhea and recurrent pelvic pain, can lead to delays in diagnosis. However, advancements in diagnostic imaging, such as 3D transvaginal ultrasound and MRI, have greatly improved our ability to identify ACUM more accurately. Surgical intervention, especially laparoscopic resection, is the most effective treatment, with careful consideration for fertility preservation in younger women. Further research, particularly through longitudinal studies, is crucial to gain a deeper understanding of ACUM's natural progression and its potential impact on reproductive health.

CLINICAL SIGNIFICANCE

The clinical significance of this study lies in its potential to bridge the gap between research findings and practical healthcare applications. It emphasizes the importance of translating scientific observations into meaningful improvements in patient care, diagnosis, and treatment outcomes. By highlighting real-world relevance, the study contributes to evidence-based medical practice and supports informed clinical decision-making. Ultimately, the findings aim to enhance patient quality of life, optimize therapeutic strategies, and promote better disease management in clinical settings.

ABBREVIATIONS

ACUM: Accessory Cavitated Uterine Malformation

MRI: Magnetic Resonance Imaging

USG: Ultrasonography CPP: Chronic Pelvic Pain LSC: Laparoscopic Surgery

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AUTHOR CONTRIBUTIONS

All authors significantly contributed to the study conception and design, data acquisition, or data analysis and interpretation. They participated in drafting the manuscript or critically revising it for important intellectual content, consented to its submission to the current journal, provided final approval for the version to be published, and accepted responsibility for all aspects of the work. Additionally, all authors meet the authorship criteria outlined by the International Committee of Medical Journal Editors (ICMJE) guidelines.

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CONFLICT OF INTEREST

Authors declared that there is no conflict of interest.

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CONSENT FOR PUBLICATION

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DATA AVAILABILITY

All data generated and analyzed are included within this research article. The datasets utilized and/or analyzed in this study can be obtained from the corresponding author upon a reasonable request.

USE OF ARTIFICIAL INTELLIGENCE (AI) & LARGE LANGUAGE MODEL (LLM)

The authors confirm that no AI & LLM tools were used in thewriting or editing of the manuscript, and no images were altered or manipulated using AI & LLM.

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