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Rare Case Of Accesory Cavitated Uterine Malformation

Dr. A. Sithi Sakhifa[1], Dr. P. Bharathi[2], Dr. R. Jayanthi[3], Dr. A. Kala[4]

[1,2,3,4]Post Graduate, Assistant Professor, HOD and Chief, Professor, Department of Obstetrics and Gynecology, Meenakshi Medical College Hospital and Research Institute, Enathur, Kanchipuram. Meenakshi Academy of Higher Education And Research(MAHER), Chennai.

Email ID: sithishakifa06@gmail.com

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ABSTRACT

Accessory Cavitated Uterine Malformation (ACUM) is a rare and often underdiagnosed congenital uterine anomaly that typically presents in adolescents and young women with severe dysmenorrhea and pelvic pain. It is characterized by the presence of an accessory, non-communicating uterine cavity lined by functional endometrium within an otherwise normal uterus. ACUM is often mistaken for other Müllerian anomalies or gynecological pathologies such as hematometra, endometriosis, or fibroids due to overlapping clinical and radiological features.

We report a rare case of a 16-year-old adolescent female who presented with severe cyclical lower abdominal pain since menarche, which was not relieved by medical management. Pelvic ultrasonography and MRI revealed a normal uterine anatomy with a well-circumscribed cystic lesion in the anterior wall of the uterus, suggestive of a cavitated lesion containing altered blood products. Diagnostic laparoscopy confirmed the presence of a non-communicating accessory cavity within the myometrium. Complete excision of the cavity and histopathological evaluation confirmed the diagnosis of ACUM.

This case highlights the importance of considering ACUM as a differential diagnosis in young females presenting with intractable dysmenorrhea, especially when imaging shows a cystic lesion within a normal uterus. Early and accurate diagnosis is essential to relieve symptoms and prevent complications such as infertility or progression to chronic pelvic pain. Surgical excision remains the treatment of choice, offering complete symptom resolution and improved quality of life. Awareness of this rare condition among clinicians and radiologists is crucial to avoid unnecessary diagnostic delays and interventions.

Keywords: *Accessory cavitated uterine malformation, adolescent dysmenorrhea, congenital uterine anomaly, diagnostic laparoscopy, Müllerian anomaly*

1. INTRODUCTION

Accessory Cavitated Uterine Malformation (ACUM) is an extremely rare congenital Müllerian anomaly that predominantly affects young, nulliparous women under the age of 30 years, typically presenting with chronic cyclic pelvic pain and severe dysmenorrhea refractory to medical management (1,2). First described as a distinct clinical entity in recent decades, ACUM is often misdiagnosed due to its rarity and overlapping clinical features with other uterine anomalies such as rudimentary horns, adenomyosis, or non-communicating uterine horns (3).

Pathologically, ACUM is defined as an isolated, cavitated, accessory mass lined by functional endometrial tissue, surrounded by a smooth muscle wall, and located subserosally beneath the insertion point of the theround ligament on the anterior uterine wall (4). Importantly, this mass does not communicate with the normal endometrial cavity or cervix, and is often associated with chocolate-brown fluid accumulation due to retained menstrual blood, mimicking endometriotic cysts (5).

Diagnosis hinges on specific criteria: (a) a single accessorycavitated mass; (b) a normal uterus with regular endometrial cavity, fallopian tubes, and ovaries; (c) histopathological confirmation of an endometrial-lined cavity with glands and stroma; (d) intraoperative evidence of a non-communicating mass; (e) presence of thick, dark fluid content; and (f) absence of adenomyosis in the surrounding myometrium (1,6).

Due to its non-specific imaging findings and unfamiliarity among clinicians, the diagnosis of ACUM is frequently delayed or misclassified, leading to prolonged patient distress and suboptimal treatment (7). Early recognition and laparoscopic excision of the lesion is considered both diagnostic and curative, with most patients reporting complete symptom resolution postoperatively (3,5).

2. CASE PRESENTATION

Patient Demographics:

A 42-year-old premenopausal female, Mrs. A, gravida 3 para 2, presented to the Obstetrics and Gynecology Outpatient Department (OG OPD) with a chief complaint of heavy menstrual bleeding (menorrhagia) persisting over the past six months. The menstrual bleeding was described as progressively worsening, often soaking through multiple pads per day and interfering with her daily activities.

In addition, she reported severe lower abdominal and pelvic pain (dysmenorrhea) during menstruation, which was not relieved by standard medical therapy, including NSAIDs and hormonal medications previously prescribed by her primary physician.

She denied any history of intermenstrual bleeding, postcoital bleeding, or vaginal discharge. There was no significant weight loss, fever, or urinary or bowel symptoms. Her obstetric history was uneventful, and she had regular antenatal check-ups during her previous pregnancies. She had no prior surgeries or known medical comorbidities.

Clinical Examination:

On general physical examination, the patient was alert and oriented, with stable vital signs. There was no pallor or signs of anemia, and systemic examination was within normal limits.

Per abdominal examination revealed no palpable mass or tenderness.

Per speculum examination showed a healthy cervix and vagina with no visible lesions or discharge.

Bimanual pelvic examination revealed a bulky, irregular uterus, approximately equivalent to a 10–12-week gravid uterus in size. The uterus was firm in consistency and mobile, with no adnexal masses or tenderness appreciated on either side.

Investigations:

Laboratory Investigations:

- Hemoglobin: 10.2 g/dL (suggestive of mild anemia)
- White blood cell count and differential: Within normal limits
- Platelet count: Normal
- Coagulation profile: Normal
- Thyroid profile: Normal
- Pap smear: Negative for intraepithelial lesion or malignancy

Imaging Studies:

1. **Pelvic Ultrasound (Transabdominal and Transvaginal):**

   The ultrasound revealed a hypoechoic lesion with cystic degeneration in the anterior myometrium, measuring approximately 5.5 x 4.8 cm. The lesion appeared to displace the endometrial stripe posteriorly. The endometrial thickness was within normal range. The lesion was suspicious for a degenerating fibroid or possibly adenomyosis, but findings were not definitive.

2. **MRI Abdomen and Pelvis:**

   To further evaluate the lesion, MRI was performed. It showed a well-circumscribed hypodense lesion located in the anterior myometrial wall with areas of cystic changes and no contrast enhancement, measuring 5.8 x 4.9 cm. The lesion was displacing the endometrial cavity posteriorly but without infiltration. There was no evidence of malignancy, lymphadenopathy, or adnexal involvement. The radiological impression was suggestive of a cystic degenerating leiomyoma.

Diagnosis:

Based on the clinical presentation, imaging studies, and lack of response to medical therapy, a provisional diagnosis of symptomatic uterine fibroid with cystic degeneration was made.

Treatment:

Given the chronic nature of the patient's symptoms, failure of conservative medical management, and the significant impact on her quality of life, a definitive surgical intervention was considered the most appropriate next step. The patient was thoroughly counseled regarding the available treatment options, including laparoscopic mass excision, myomectomy, and hysterectomy. In view of her completed family, desire for definitive symptom relief, and preference to avoid recurrence, she

opted for total removal of the uterus and adnexa.

The patient subsequently underwent an elective Total Abdominal Hysterectomy with Bilateral Salpingo-Oophorectomy (TAH + BSO) under general anesthesia. The procedure was uneventful, and the patient had a satisfactory postoperative recovery.

Surgical intervention is widely regarded as the mainstay of treatment for ACUM, particularly in symptomatic individuals who have not responded to medical therapies. The ideal surgical approach, especially in younger women desiring fertility preservation, is laparoscopic excision of the accessory cavitated mass, which enables complete lesion removal while preserving the uterus. This approach significantly improves both symptoms and reproductive outcomes.

On the other hand, hormonal treatments such as combined oral contraceptives (OCPs), GnRH agonists, and levonorgestrel intrauterine systems (LNG-IUD) may offer temporary symptomatic relief. However, these methods do not address the structural abnormality, and symptoms often recur following cessation of therapy. Ethanol sclerotherapy has also been explored in a limited number of cases for women seeking uterine conservation, though results have been variable and long-term efficacy remains unclear.

Overall, current evidence supports that hormonal therapy alone is less effective in providing sustained symptom relief and often requires prolonged follow-up. In contrast, surgical management offers definitive resolution, especially when appropriately individualized based on the patient's age, reproductive plans, and symptom burden.

Intraoperative Findings:

- A bulky uterus with a 6 cm intramural fibroid located in the anterior wall with cystic areas on the surface
- Normal bilateral ovaries and fallopian tubes
- No adhesions or evidence of pelvic endometriosis
- No ascites or abnormal peritoneal deposits

The surgery was completed uneventfully with minimal blood loss. The excised specimen was sent for histopathological examination.

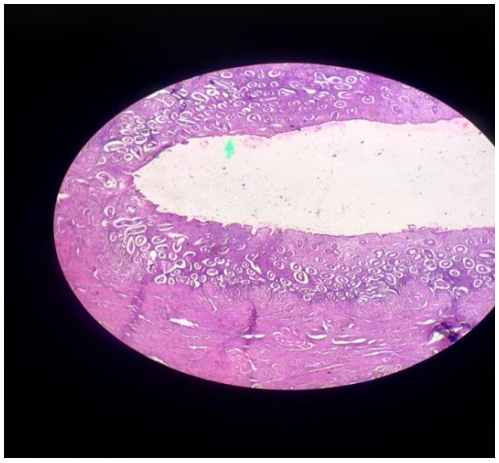
Histopathological Findings:

- The uterine specimen showed a well-encapsulated intramural leiomyoma with areas of hyaline and cystic degeneration
- No evidence of atypia, necrosis, or malignancy
- Endometrium was proliferative in nature
- Ovarian and tubal tissues were unremarkable

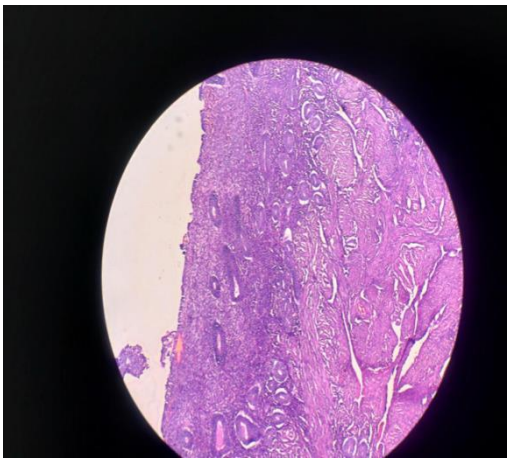
The definitive diagnosis of Accessory Cavitated Uterine Mass (ACUM) is established through histopathological examination (HPE). The typical histological findings include an accessory cavity lesion located adjacent to the myometrium, which is non-communicating with the endometrial cavity. The cystic cavity is lined by functional endometrial tissue, comprising endometrial glands and stroma embedded within hypertrophic myometrium. These features distinguish ACUM from adenomyosis, endometriosis, and congenital anomalies such as rudimentary uterine horns.

–Histopathological Findings –

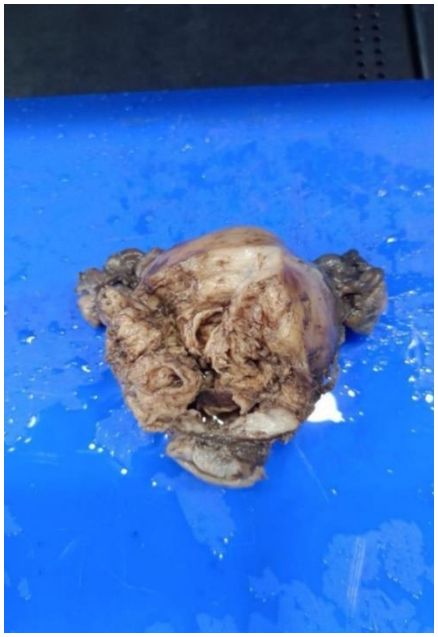
Figure 2



Histopathological Findings- Figure1



Histopathological Findings- Figure2



Gross finding - Figure 3

Gross finding -Figure 4

Postoperative Course and Outcome:

The patient had an uneventful postoperative recovery. She was started on oral fluids within 12 hours post-surgery and gradually progressed to a full diet. Intravenous analgesics and antibiotics were administered as per protocol. She was mobilized early and discharged on the fourth postoperative day.

At her two-week, three-month, and six-month follow-up visits, the patient remained asymptomatic. She reported complete relief from her previous symptoms, no urinary or bowel complaints, and no menopausal symptoms during this period. She was advised routine follow-up every six months and general health surveillance.

Prognosis: The prognosis of ACUM is favorable, especially with early diagnosis and timely surgical treatment. Most patients experience complete or near-complete symptom relief following excision. Studies show that up to 90% of patients achieve long-term symptom resolution post-surgery.

Patients managed with medical therapy alone may require prolonged treatment and follow-up, with a higher risk of symptom recurrence once therapy is stopped. Thus, ongoing clinical monitoring is crucial in these cases.

3. DISCUSSION

Accessory Cavitated Uterine Mass (ACUM), previously referred to as juvenile cystic adenomyoma, is a rare and often underdiagnosed Müllerian anomaly characterized by an isolated, cavitated, non-communicating lesion within the myometrium, typically near the uterine horn. First recognized as a distinct clinical entity in the early 2000s, ACUM primarily

affects young women and adolescents, most of whom present with severe, progressive dysmenorrhea and chronic pelvic pain from the time of menarche.

The defining pathological hallmark of ACUM is a cystic lesion lined by functional endometrial glands and stroma, surrounded by hypertrophic myometrial smooth muscle. The cyst contains hemorrhagic or chocolate-colored menstrual fluid, which accumulates cyclically due to the lack of communication with the endometrial cavity. This pathophysiology explains the intensity and cyclical nature of the pain, often mimicking other gynecologic conditions.

Diagnostic Challenges and Imaging

Diagnosis of ACUM is notably difficult due to its non-specific imaging findings and overlapping features with more common conditions such as: (differential diagnosis)

- Adenomyosis
- Endometriosis
- Cystic degeneration in fibroids
- Unicornuate uterus with obstructed rudimentary horn
- Rudimentary uterine horn

Ultrasound typically reveals a cystic or hypoechoic lesion within the myometrium, but fails to clearly define its origin or nature. MRI offers superior soft tissue characterization and may demonstrate a well-circumscribed, intramural cystic lesion, with high T1 and T2 signals (due to blood products), and absence of connection to the endometrial cavity. However, as noted by Takeuchi et al. (2010), even MRI cannot definitively differentiate ACUM from similar entities without histopathological confirmation.

Comparison with Other Studies

In a retrospective study by Acién et al. (2012), who first systematically described and named ACUM, 30 young women with severe dysmenorrhea were reviewed. They found that surgical excision of the lesion resulted in complete symptom resolution, supporting the theory that ACUM is a developmental Müllerian anomaly and not a variant of adenomyosis.

Similarly, Batt et al. (2011) emphasized that the key differentiator of ACUM from adenomyosis lies in its focal and encapsulated structure, and the younger age group of presentation, in contrast to the diffuse nature and later onset of adenomyosis in women aged 35 years and above.

A study by Yuan et al. (2016) involving adolescent patients with ACUM also highlighted the high rate of misdiagnosis, with several patients being treated for presumed endometriosis or fibroids until definitive surgical excision and histology revealed ACUM. This study reinforced the need for high clinical suspicion when encountering young women with intractable dysmenorrhea unresponsive to medical management.

Management Approaches

Management of ACUM varies depending on patient age, reproductive desire, symptom severity, and lesion size. Conservative therapies like NSAIDs, combined oral contraceptives (OCPs), Levonorgestrel-releasing intrauterine devices (LNG-IUDs), and GnRH agonists may offer temporary symptom relief but often fail to address the underlying structural anomaly.

Definitive treatment involves surgical excision of the mass, preferably via laparoscopy, which allows for precise removal while preserving uterine integrity, particularly important for women desiring future fertility. In cases where the lesion is large, recurrent, or where fertility is no longer a concern, hysterectomy may be considered.

Numerous reports, including one by Fedele et al. (2005), demonstrate that complete surgical removal of the ACUM leads to dramatic symptom improvement, with most patients becoming pain-free and resuming normal activities postoperatively.

Conclusion

ACUM remains a clinically significant but underrecognized cause of severe dysmenorrhea in young women. The lack of specific imaging characteristics, combined with overlapping features with other gynecological disorders, makes diagnosis challenging. A high index of suspicion, especially in young females with recurrent pelvic pain unresponsive to analgesics, is crucial. MRI, while helpful, may not always be conclusive; thus, laparoscopic evaluation and surgical excision remain the gold standard for both diagnosis and treatment. Comparative studies strongly support surgical intervention as the most effective approach, with excellent long-term outcomes and symptom resolution.

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