



CASE REPORT

Accessory Cavitated Uterine Mass Masquerading as Endometriotic Cyst, in a Multiparous Woman with Refractory Dysmenorrhea

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Introduction

Accessory Cavitated Uterine Mass (ACUM) is a rare uterine anomaly comprising of an accessory uterus-like mass in the uterus along the wall but with no communication to the main uterus and having a functional endometrium. ACUM is commonly misdiagnosed, as the awareness about this entity is low [1]. Mislabeling ACUM as endometriotic cyst is much rarer [2]. Severe dysmenorrhea, not responding to medical management, is the usual presenting complaint. Although more commonly reported in young nulliparas, the characteristic ultrasound features should evoke suspicion even in multiparas [1].

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Case Report

A 32 year-old multiparous lady presented to us with complaints of severe, progressive dysmenorrhea and chronic pelvic pain for last 4 years. She attained menarche at 13 years of age, following which she had regular painless cycles for 10 years. She had spontaneous conceptions with two vaginal deliveries, children now aged 14 and 11 years. There was history of two medical abortions, followed by dilation and curettage of third unwanted pregnancy along with laparoscopic tubal ligation at a local hospital, eight years back. She started to experience severe lower abdominal pain, which would start 10 days prior to menstruation and increase remarkably during menstruation for last four years. The pain progressively increased over time to an extent that she only had 5–6 pain-free days per cycle, requiring emergency admissions and intravenous analgesics up to ten times a year. She also developed dyspareunia that was contributory to her separation from husband.

At first evaluation at local hospital, ultrasound pelvis reported it as “thick-walled cyst in left adnexa, closely abutting left ovary with heteroechoic contents, measuring 2.0 × 1.9 cm, likely to be endometriotic cyst.” Her records revealed at least eight ultrasound reports with similar findings and a final impression of left endometriotic cyst. Only one ultrasonologist had kept a differential diagnosis of subserosal fibroid with degeneration. CA-125 ranged from 46.2 U/mL to 57.05 U/mL. She was advised 3 courses of 3 monthly dinogest 2 mg once daily at various hospitals. She even tried alternative medicine, but got no relief. She was referred to a tertiary center, where she was advised oral contraceptive pills (Ethinyl Estradiol 30ug + Levonorgestrel 150 ug) for 3 months, without any alleviation of pain. Considering her young age and small size of lesion, panhysterectomy had not been offered to her. With unsatisfactory outcome and fear of recurrence, she kept on consulting several doctors.

At our center, on bimanual examination the fundus of uterus was felt toward the right side, while a globular mass

of approximately 3 cm size was felt anteriorly and towards the left side with free fornices. Confirmatory transvaginal ultrasound (4–9 MHz endocavitary probe of Phillips Clear-Vue 350) done by us revealed normal size and location of bilateral ovaries. These findings raised suspicion to reconsider the original diagnosis. A thick-walled cystic lesion of approximately 2.5 × 2.5 × 2.9 cm was seen adjacent to the uterus, toward the left side, with normal delineation of the rest of the uterus and endometrial cavity. Findings were confirmed by the radiologist on 2D/3D ultrasound, and a diagnosis of Accessory Cavitated Uterine Mass was made. (Fig. 1).

At laparotomy, a bulge was noted on the uterus, between left cornua and round ligament insertion. Vasopressin was injected on the surface of the bulge. After incising the myometrium, chocolate fluid was drained from the thick-walled cavity. Cyst wall was separated from myometrium with use of knife plus monopolar cautery, and base was clamped and cut. (Fig. 2) No.1 braided polyglactin suture was used to

close the uterine incision. Both ovaries were noted to be of normal size and shape, while Fallope’s rings were seen on medial stumps of fallopian tubes as well as one on the left round ligament. Histopathology reported an endometrial lined cavity with smooth muscle in the wall, consistent with ACUM. A month post-surgery, she is now asymptomatic, after having experienced years of worsening pelvic pain and analgesic dependence.

Discussion

Accessory Cavitated Uterine Mass is a developmental anomaly of the Mullerian system, which can cause significant distress to the patient. The delay in arriving at the correct diagnosis further adds to the trauma. Even though more than 60 cases have been reported till date, including the first probable case reported way back in 1912, general awareness remains low.

Fig. 1 Ultrasound images **1a**: 2D ultrasound image showing a thick-walled cyst with ground glass content **1b**: 3D ultrasound image showing a thick-walled cyst from left lateral wall of uterus, with normal uterine cavity suggestive of ACUM

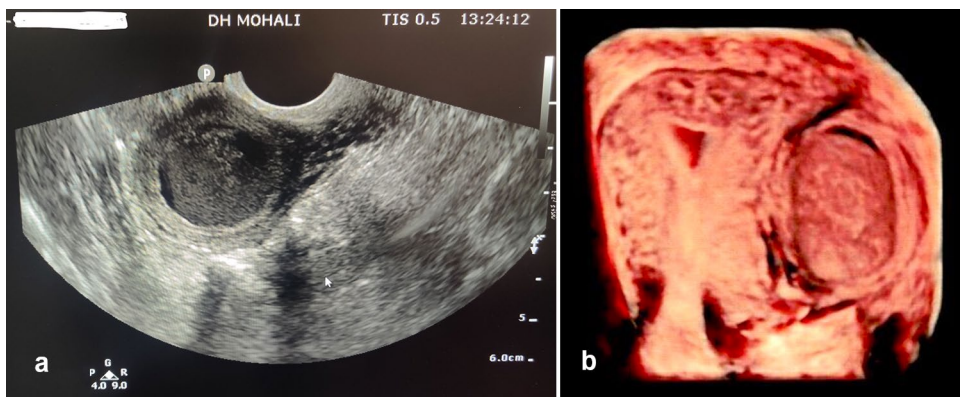
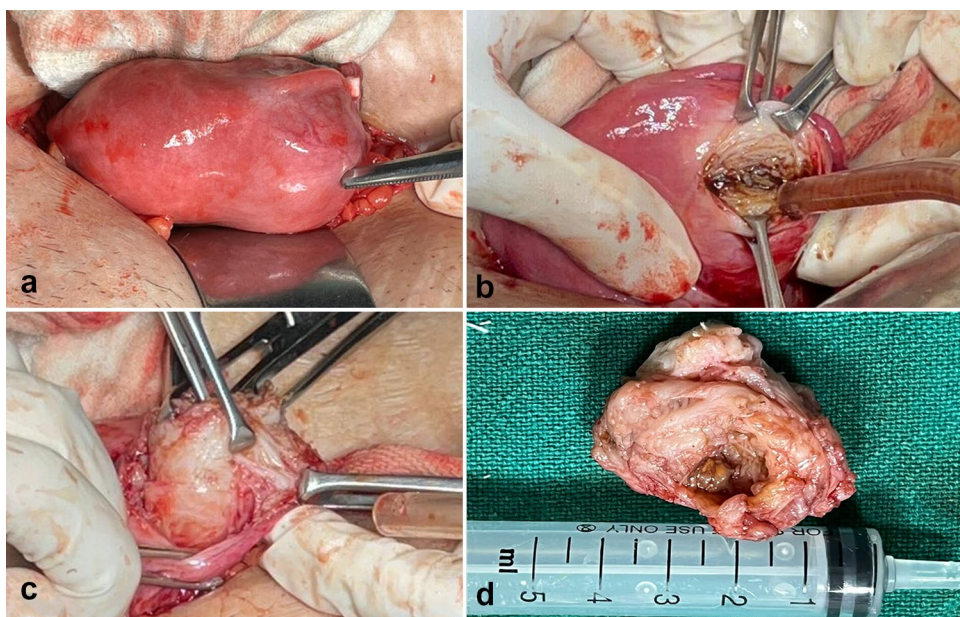


Fig. 2 Steps of surgical procedure and specimen **2a**: Bulge seen on uterus at the origin of left round ligament, **2b**: Uterine incision over the lesion drained chocolate material, **2c**: Excision of the cyst wall from myometrium, **2d**: surgical specimen of ACUM



ACUM is classified as a non-communicating variety of uterine-like masses (ULM). Possible embryological basis has been hypothesized as persistence or duplication of Mullerian tissue due to gubernaculum dysfunction, since the usual location of the lesion is at the origin of one of the round ligaments [3].

Majority of cases have been seen in young, nulliparous women, with less than 10% reported in women older than 30 years [4]. Our patient was a multipara aged 32 years, traditionally considered a low-risk candidate for developmental uterine anomaly. Misdiagnosis is common, with cases having been missed even at surgery. Azuma et al. [5] have published a case of 37 year-old women with dysmenorrhea, who had undergone endometriotic cystectomy at age of 30 years, followed by conception and delivery, with recurrence of symptoms as soon as she resumed menstruation. She was medically managed with presumption of diagnosis of recurrence of endometriosis, but did not respond, just like our case. Although a transvaginal ultrasound suggested a fibroid like 27 mm mass and MRI revealed a cavity with blood component adjacent to uterus, a definite preoperative diagnosis of ACUM was not made. She opted for total laparoscopic hysterectomy, instead of uterine preserving surgery. Our patient also changed five hospitals, including taking trial of alternative medicine with multiple visits to emergency department, in her quest for relief from the debilitating pain. Unfortunately, the lesion was not detected even at the time of laparoscopic tubal ligation and we believe that the round ligament of left side had been mistakenly ligated, due to probable distortion of anatomy.

Numerous ultrasounds had given the probable diagnosis of endometrioma, even though the detailed description of the lesion mentioned that a blood-filled cavity was seen abutting the left ovary and not part of the ovary. We wish to highlight the features on transvaginal ultrasound which should raise suspicion of ACUM—a normal uterine cavity, an accessory well-defined globular blood-filled cavity with surrounding myometrium (Both differentiating from Rudimentary horn) and the presence of separate bilateral normal ovaries (differentiating feature from endometrioma). 3D ultrasound has an essential role in confirming the diagnosis and proving the presence of normal uterine cavity, thus obviating the need for MRI, hysterosalpingography and also hysteroscopy [6].

Medical management is ineffective. Surgical excision via laparoscopy, laparotomy or robot-assisted techniques are gratifying as the patients experience pain relief from the very next menstrual cycle. One month postoperatively, our patient is significantly relieved of symptoms, for the first time in years.

Conclusion

Accessory Cavitated Uterine Mass is an easily treatable cause of refractory dysmenorrhea, which although commoner in young, nulliparous women, can also be seen in multiparas more than 30 years of age. Ultrasound features of normal uterus, adjacent endometrioma-like blood-filled cavity with surrounding thick myometrium and normal bilateral ovaries, should raise suspicion. Early diagnosis can save the patient from losing valuable years to low quality of life and analgesic dependence.

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Declarations

Conflict of Interest Japleen Kaur, Shikha Rani, Ravneet Kaur Gill, Sushmita Sharma, Nisha Bhagat declare that they have no conflict of interest.

Ethical Approval Not applicable as patient was managed according to standard treatment and no experimental intervention was done.

Informed Consent Written Informed consent was taken from the patient.

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