

# Accessory cavitated uterine mass (ACUM) as a miniature uterine anomaly causing severe lateralized dysmenorrhea: case series

Weronika Zajczkowska<sup>1</sup> , Karina Kapczuk<sup>1,2</sup> 

<sup>1</sup>Gynecology and Obstetrics Clinical Hospital of Poznan University of Medical Sciences, Poland

<sup>2</sup>Division of Gynecology, Poznan University of Medical Sciences, Poland

## ABSTRACT

**Objectives:** Our study aimed to retrospectively analyse and present the clinical course of accessory cavitated uterine mass (ACUM), a rarely diagnosed uterine malformation.

**Material and methods:** The study group comprised 5 adolescents and young adult women that were treated in the Division of Gynecology, Clinical Hospital of Obstetrics and Gynecology of Poznan University of Medical Sciences, between October 2017 and August 2022. Patients' age at diagnosis of ACUM ranged from 14.1 to 27.5 (mean 21.4) years. All patients complained of severe dysmenorrhea with significant lateralisation of the pain.

**Results:** Pelvic ultrasound (US) followed by pelvic magnetic resonance imaging (MRI) revealed the presence of a small cystic lesion surrounded by a ring of myometrium within or in connection with the regular uterine body. In four patients (80%), the lesion was on the right side, and in one patient (20%) on the left side. The volume of the ACUM cavity ranged from 0.04 to 2.4 (mean 0.8) cm<sup>3</sup>. Laparoscopic excision of ACUM, located near the uterine attachment of the round ligament, was performed in all five cases and resulted in the complete resolution of symptoms. None of the patients was diagnosed with adenomyosis or pelvic endometriosis.

**Conclusions:** ACUM is a small, surgically correctable cause of severe dysmenorrhea in young females with an otherwise normal uterus. The lateralisation of the menstrual pain should prompt the search for this malformation with imaging techniques (US, MRI). ACUM laparoscopic excision results in complete relief of symptoms. ACUM is not associated with pelvic endometriosis.

**Keywords:** accessory cavitated uterine mass; accessory cavitated uterine malformation; ACUM; dysmenorrhea

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## INTRODUCTION

Dysmenorrhea is painful menstruation that might co-occur with nausea, vomiting or diarrhea. In primary dysmenorrhea, pelvic pathologic conditions are absent. It usually presents after the onset of ovulatory cycles. Secondary dysmenorrhea is associated with pathological conditions of the pelvic organs. The most common cause of secondary dysmenorrhea is endometriosis. Dysmenorrhea that occurs with or early after menarche may indicate an obstructive Müllerian anomaly as a cause [1]. Approximately 7% of young women have an anatomical abnormality in their reproductive tract; the most frequent symptom is recurrent, severe pelvic pain [2].

Accessory cavitated uterine mass (ACUM) or accessory cavitated uterine malformation (ACUM) is a rare unclassified uterine anomaly [class U6 according to the European Society of Human Reproduction and Embryology and the European Society for Gynecological Endoscopy (ESHRE/ESGE) classification system of female genital tract congenital anomalies [3], variant according to the American Society for Reproductive Medicine Müllerian anomalies classification (ASRM MAC) 2021] [4]. The lesion has been described in the literature with different terminologies: juvenile cystic adenomyoma, isolated cystic adenomyoma, cavitated adenomyoma, accessory cavitated masses, and uterine-like mass until Ación et al. [5] termed these lesions as ACUM in 2012.

### Corresponding author:

Weronika Zajczkowska

Gynecology and Obstetrics Clinical Hospital of Poznan University of Medical Sciences, 33 Polna St, 60–535 Poznan, Poland

e-mail: zajczkowskaweronika@gmail.com

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Accessory cavitated uterine mass is an isolated non-communicating uterine lesion with functional endometrium that lies within the normally shaped and functioning uterus and is most often located close to the uterine insertion of the round ligament [6]. It is considered a congenital Müllerian anomaly caused by ectopia or by the duplication and persistence of the ductal Müllerian tissue (displaced tissue) at the round ligament attachment level. It might be related to gubernaculum dysfunction [7].

Accessory cavitated uterine mass is usually diagnosed in menstruating girls and young women with severe and recurrent perimenstrual pelvic pain, resistant to treatment with non-steroidal anti-inflammatory drugs (NSAID) or oral contraceptive pills (OCP). The diagnostic criteria proposed by Acién et al. [6] to diagnose ACUM are as follows: (1) an isolated, accessory, cavitated mass; (2) a normal uterus (with endometrial lumen), fallopian tubes and ovaries; (3) surgical evidence with an excised mass and pathological finding; (4) an accessory cavity lined by endometrial epithelium, with glands and stroma; (5) chocolate-brown-colored fluid content; and (6) no adenomyosis. Obstructive uterine horns, adenomyosis and degenerating fibroids are pathological conditions of the uterus to consider in the differential diagnosis [5]. Non-invasive imaging modalities that help make an accurate preoperative diagnosis are ultrasound (US) and magnetic resonance imaging (MRI). According to Gupta et al. [8], the pelvic US criteria to establish the correct diagnosis of ACUM are: (1) site of the lesion along the presumed location of the round ligament, (2) characteristic appearance of the lesion: echotexture similar to uterine myometrium with central contents showing endometrioma-like ground glass appearance and lining of the cavity similar to uterine endometrium, (3) documentation of vascularity in the lesion, (4) appearance of bilateral normal uterine cornua (on 3D US) and normal ovaries, (5) normal appearance of the rest of the uterus with no features suggestive of adenomyosis. The MRI findings in ACUM are a well-defined, cavitated uterine wall mass located immediately below the usual round ligament insertion site. Normal central T2 hyperintense endometrial lining shows hemorrhagic contents within, which appear hyperintense on T1WI with no signal suppression on fat-suppressed images and reflect hypointense shading on T2WI. The wall of the cavity is hypointense on T2WI and comprises myometrial tissue. Besides, the uterine cavity is normal with normal bilateral cornua [8]. The normal endometrial cavity has no communication with the accessory cavity [9].

The true prevalence of ACUM is yet to be established due to the low number of diagnosed cases and the condition's rarity [10].

Our study aimed to retrospectively analyze and present the clinical course of ACUM in adolescents and young adult women diagnosed and treated in our center.

## MATERIAL AND METHODS

The presented series of cases involves five patients diagnosed with ACUM. The patients were treated in the Division of Gynecology, Gynecology and Obstetrics Clinical Hospital of Poznan University of Medical Sciences (tertiary care hospital), Poland, between October 2017 and August 2022. Preoperative evaluation of each patient involved: clinical examination, pelvic US and pelvic MRI. We have analyzed the patients' medical records and assessed the following data: age at diagnosis; presentation; preliminary diagnosis; concomitant diseases; location and size of ACUM; performed surgery; histopathologic diagnosis; outcomes.

Following our institution's ethics guidelines, approval to conduct this study was not required. However, we informed all patients or their parents, who signed informed consent forms before the surgery, that their clinical data might be used for research and scientific publication.

## RESULTS

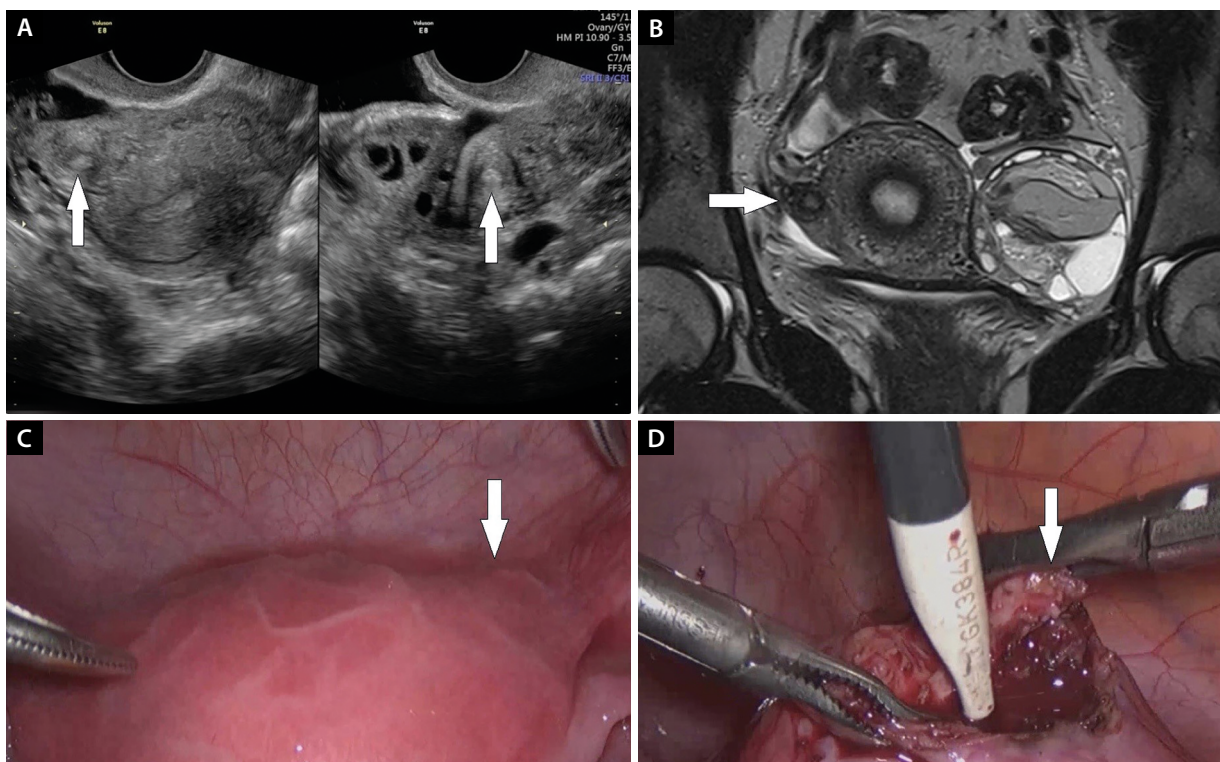
In our subjects (Tab. 1), the age at diagnosis of ACUM ranged from 14.1 to 27.5 years (mean 21.4 years). All patients complained about severe dysmenorrhea with distinct lateralization of the pain in the lower abdomen: in four patients (80%) on the right side and in one patient (20%) on the left side. None of these patients was pregnant before. We admitted patient 1 to our unit with a preliminary diagnosis of a unicornuate uterus with a non-communicating functional horn with a rudimentary cavity. Patients 2 and 3 were admitted with a preliminary diagnosis of endometriosis and necrotic uterine leiomyoma, respectively. Patient 4 was consulted for dysmenorrhea. A left ovarian mass (mature teratoma 5cm in diameter) was found, but the tumor's location was contralateral to the side of the pain, which the patient suffered. Patient 5 was consulted for dysmenorrhea and a cystic uterine lesion found on the abdominal US.

Pelvic US followed by pelvic MRI revealed normal uterine anatomy and the presence of a small cystic lesion surrounded by a ring of myometrium, which was located within the myometrium (patients 1, 4 and 5) (Fig. 1) or connected with the outer myometrium (patients 2 and 3) (Fig. 2) of the upper part of the anterior wall of the uterine body near the uterine horn. The mean volume of ACUMs cavity, calculated with the ellipsoid formula, was  $0.8 \text{ cm}^3$  (range  $0.04\text{--}2.4 \text{ cm}^3$ ). The lesion was on the right side in 4/5 (80%) of our patients. In one patient (20%), it was located on the left side. Laparoscopic excision of ACUM (the lining of the cavity and the myometrial ring) was performed in all five cases. In patients 2 and 3, the lack of connection of ACUM with the ipsilateral fallopian tube was confirmed with intrauterine blue methylene injection at the beginning of laparoscopy. The laparoscopic

**Table 1.** Characteristics of our five patients diagnosed with accessory cavitated uterine mass (ACUM)

N	Age at diagnosis [years]	Preliminary diagnosis	Coexisting disease	ACUM location	ACUM cavity volume [cm <sup>3</sup> ]
1	14.7	Unicornuate uterus + functional horn non-communicating	None	R within the myometrium	1.0
2	24.6	Endometriosis	None	R in connection with the outer myometrium and within the broad ligament	0.2
3	20.2	Uterine leiomyoma	None	R in connection with the outer myometrium, mainly within the broad ligament	2.4
4	27.5	Dysmenorrhea left, ovarian mass	Autoimmune thyroiditis	R in the outer myometrium	0.04
5	14.1	Dysmenorrhea, cystic uterine lesion	Congenital hypothyroidism, Left ear hearing loss	L within the myometrium	0.2

R — right; L — left

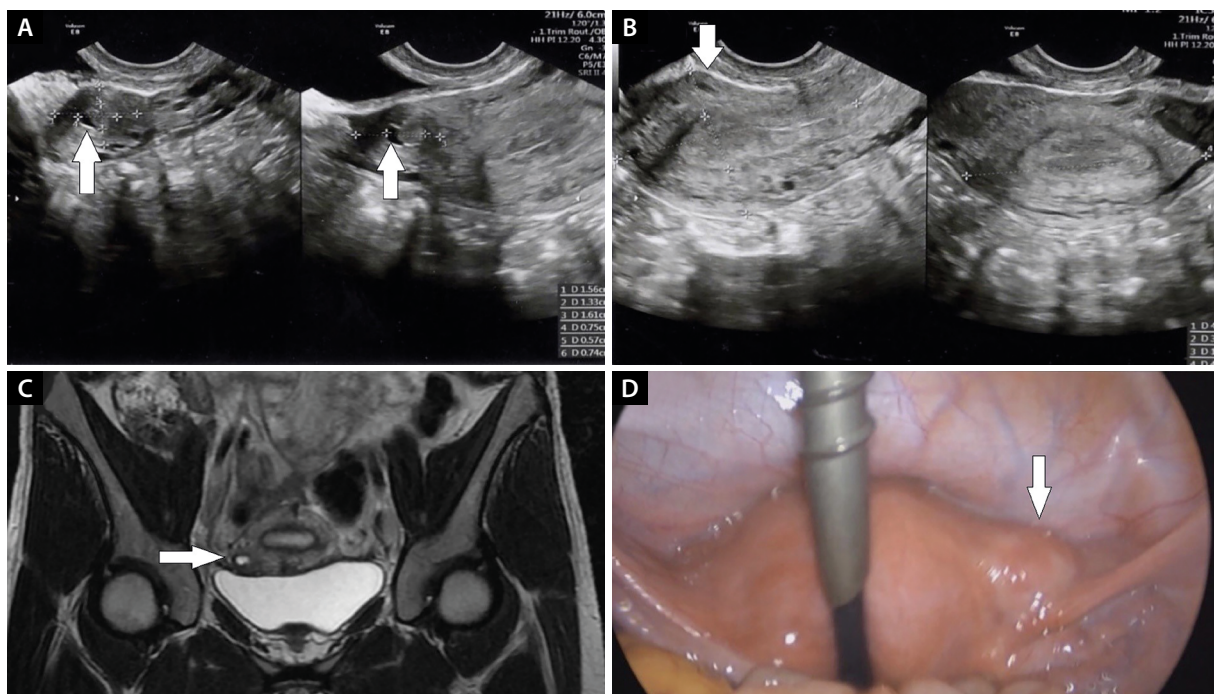


**Figure 1.** Patient 4; **A.** Transvaginal pelvic ultrasound (US) showing cavitated mass surrounded by a ring of the myometrium, located in the outer myometrium on the right side; **B.** Magnetic resonance imaging (MRI) showing cavitated, uterine wall mass with T2 hyperintense endometrial lining; **C.** Normal appearance of the uterus during laparoscopy. Accessory cavitated uterine mass (ACUM) is located within the outer myometrium next to the uterine attachment of the right round ligament; **D.** Laparoscopic excision of the mass filled with a thick, brownish fluid

appearance of the uterus in patients 4 and 5 (Fig. 1) was normal, making ACUM identification and excision more challenging. In each patient, the excised mass was filled with a thick, brownish fluid, and a histopathological examination confirmed the preoperative diagnosis of ACUM.

None of our patients was diagnosed with adenomyosis or pelvic endometriosis. Laparoscopic excision of ACUM resulted in the resolution of the pelvic pain in all the subjects (the patients were followed up 3 and 6 months after surgery).





**Figure 2.** Patient 2; **A, B.** Transvaginal pelvic ultrasound (US) showing cavitated mass surrounded by a ring of the myometrium, connected with the outer myometrium of the upper part of the anterior uterine wall and otherwise normal uterus; **C.** Magnetic resonance imaging (MRI) showing cavitated uterine wall mass with T2 hyperintense endometrial lining in connection with the outer myometrium; **D.** Accessory cavitated uterine mass (ACUM) located within the broad ligament near to the uterine attachment of the right round ligament

## DISCUSSION

The presentation of ACUM is similar to other causes of secondary dysmenorrhea, mainly endometriosis and unilateral or partial obstructive Müllerian anomalies. Severe menstrual pain in patients with ACUM is due to distention of the cavity caused by repeated bleeding within the ACUM during the menstrual periods. Based on our case series, we can assume that lateralization of the pain is a typical characteristic of menstrual pain in adolescents and young adult women with ACUM. All our patients complained about severe pain on the side of the ACUM location. In our opinion, lateralization of the menstrual pain should prompt the search for ACUM in adolescents and young adult women in which obstructed uterine horn, Robert's uterus, unilateral cervical atresia and obstructed hemivagina were excluded, and uterovaginal anatomy seemed to be normal.

We emphasize the necessity to search for ACUM as the lesion might be very small. In four of our five (80%) patients, the cavity volume within ACUM was no more than 1 cm<sup>3</sup>. Besides, in two of our patients, the lesions were both small and located mainly within the myometrium, making the laparoscopic identification of ACUM challenging. It is necessary to emphasize that the lack of preoperative diagnosis based on imaging (MRI) can result in negative laparoscopy in patients with secondary dysmenorrhea. According to Acien et al. [5], ACUMs seem larger when the woman is older. However, we

did not observe such a tendency in our case series. Pelvic MRI protocol classically recommended in uterine malformation is helpful to diagnose or rule out adenomyosis and endometriosis and to identify ACUM [11]. None of our patients was diagnosed with pelvic endometriosis, although there have been reported cases in the literature showing superficial pelvic endometriosis without evidence of adenomyosis in patients with ACUM [9–11]. Patients with severe, lateralized dysmenorrhea, even with a normal appearance of the uterus in the US, should have MRI performed and be evaluated for ACUM before a potential laparoscopy. Many reported cases of ACUM in the literature were overlooked and misdiagnosed during diagnostic laparoscopies due to its small size and lack of knowledge about this condition [6, 12, 13]. Therefore, US and MRI are necessary for the correct diagnosis preoperatively. Clinical awareness is the most crucial in establishing a proper preoperative diagnosis.

Previous studies have suggested that invasive imaging modalities such as hysterosalpingography (HSG), hysterosalpingocontrastsonography (HyCoSy) and saline infusion sonohysterography (SIS) are necessary to rule out communication between the normal endometrial cavity and the accessory cavity and thus confirm the diagnosis of ACUM [5, 6, 14, 15]. However, performing these procedures in non-sexually active adolescents or young adult women may be troublesome. Therefore, these procedures and routinely

performed hysteroscopies are unnecessary to diagnose or treat ACUM. In doubtful cases, a blue methylene injection during laparoscopy might be helpful.

As the literature states, laparoscopic excision of ACUM is the most common and effective treatment method [14–16]. In contrast to the lesions located mainly along the round ligament or within the broad ligament, for which the surgery is relatively simple, laparoscopic excision of small ACUM located within the myometrium is usually more challenging due to the difficulty in localising the cavity. ACUMs are typically located close to the uterine attachment of the round ligament. Nevertheless, it is necessary to precisely establish the location of the ACUM in relation to the uterine myometrium before the surgery. Estimating the penetration depth in the myometrium is essential to remove the lesion. In such cases, intraoperative US might also be helpful. Accurate surgical management reduces the risk of scarring of the uterus and uterine rupture in future pregnancies. ACUM laparoscopic excision results in complete relief of symptoms and minimal probability of recurrence.

Sclerotherapy is yet another reported therapeutic option. In 2020 Merviel et al. [17] presented the first published case of ethanol sclerotherapy of ACUM. The procedure involved aspiration of the fluid from the ACUM cavity and subsequent injection of the same volume of 96% ethanol for 15 minutes. Symptoms had not recurred during the two years of follow-up. In the report by Naftalin et al. [18], four cases of transvaginal ultrasound-guided alcohol sclerotherapy were presented. In one of these patients, symptoms returned six months after sclerotherapy, and the patient required laparoscopic excision. In the report by Merviel et al. [17], the diameter of the ACUM lumen was 10mm, and the volume of the collected chocolate-brown liquid was 5 cm<sup>3</sup>. In the series published by Naftalin et al. [18], the mean internal cavity diameter of the ACUM was 14 mm. In our case series, in only one patient, the mean cavity diameter of the ACUM exceeded 10mm. Therefore, transvaginal sclerotherapy with an injection of a sclerosant chemical into the lumen of the ACUM may not apply to very small ACUMs, which are more typical for adolescents. In such cases, laparoscopic ultrasound-guided sclerosant injection could be another option. Nevertheless, further studies are necessary to evaluate the long-term effectiveness of sclerotherapy of ACUM.

## CONCLUSIONS

Accessory cavitated uterine mass is a small, surgically correctable cause of severe dysmenorrhea in young females with an otherwise normal uterus. The lateralization of the menstrual pain should prompt the search for this malformation with imaging techniques (US, MRI). ACUM laparoscopic excision results in complete relief of symptoms. Accessory

cavitated uterine mass is not associated with pelvic endometriosis.

## Article information and declarations

### Ethics statement

In accordance with our institution's ethics guidelines, approval to conduct this study was not required (retrospective analysis of medical records).

### Author contributions

W.Z.: Conceptualization, data analysis, writing (original draft preparation), K.K.: conceptualization, data acquisition, writing (original draft preparation and final revision). The authors have read and agreed to the published version of the manuscript.

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### Conflict of interests

All authors declare no conflict of interest.

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