Accessory and Cavitated Uterine Mass (ACUM) in an 18-Year-Old Woman: A Case Report and Literature Review

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Case report

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Abstract

Background

Accessory and cavitated uterine mass (ACUM) is a rare uterine anomaly newly recognized as a form of developmental Mullerian anomaly, which represents a non-communicating uterus-like mass within an otherwise normal uterus. It is a benign gynecological disease associated with severe dysmenorrhea and chronic pelvic pain, which is most common in young nullipara women, and sometimes develops in parous women. Clinical manifestations combined with imaging examinations including ultrasonography (USG), magnetic resonance imaging (MRI), and hysterosalpingography (HSG) are the means to establish a correct diagnosis. Medical therapy is only marginally effective, but laparoscopic surgery for complete mass excision is a feasible technique to relieve patient's symptoms. Our article is aimed to report a case of ACUM in an 18-year-old woman and summarize the diagnostic criteria of ACUM.

Case presentation

: An 18-year-old woman was admitted for severe pain in the right lower abdomen during menstruation, which lasted more than 1 year. The patient was misdiagnosed with focal adenomyosis at our hospital on March 4, 2021. After 4 months, she was diagnosed with ACUM. Once diagnosis as focal adenomyosis, nonsteroidal anti-inflammatory drugs (NSAIDs) and gestrinone were administered to the patient. Following the diagnosis of ACUM, she received laparoscopic surgery. Our follow-up indicated that the symptom was significantly relief without drug therapy after sixty days postoperatively.

Conclusions

Clinical manifestations and imaging examinations are used to establish the diagnosis of ACUM. Medical therapy is only marginally effective, but laparoscopic surgery for complete mass excision is a feasible technique to solve the pain symptom. The prevalence and pathogenesis of ACUM and its reproductive outcomes on patients remain unclear, which calls for more and deeper research to study.

1. Background

ACUM is a rare benign gynecological disease, which is newly recognized as a form of uterine anomaly related to the malformation of Mullerian duct, and is reported that most common in nulligravid females younger than 30 years old[1, 2]. The most striking clinical feature of ACUM is early-onset, severe, progressive, and drug-resistant dysmenorrhea, which usually occurred soon after menarche[3]. The ACUM-associated dysmenorrhea was refractory to nonsteroidal anti-inflammatory drugs (NSAIDs), oral contraceptive pills, GnRHa, and analgesic drugs, while surgical resection of the lesion is the only radical treatment at present[4]. Several case reports had demonstrated the feasibility of minimally invasive laparoscopic resection for ACUM[7–9]. It is a well-circumscribed mass with a cystic cavity filled with hematometra within the myometrium adjacent to the uterine horn on imaging tests. Histopathology reveals that the wall of the accessory cavity is lined with functional endometrial glands and stroma surrounded by irregularly arranged smooth muscle[5]. Because of the lack of specificity of the imaging findings, the ACUM is often misdiagnosed as non-communicating rudimentary horn of uterus, but HSG can be used to distinguish between the two.

The pathogenesis of the disease is still controversial. Although various scholars have proposed different theories, the mainstream opinion is that ACUM suggests a new type of Mullerian anomaly[6]. Due to the published literature about ACUM consisting entirely of case reports and case reviews without population-based studies and long-term follow-up, the prevalence and long-term influences of ACUM are still unclear. There were forty-two cases of ACUM reported around the world, of which only fourteen cases provided a result of the patient's CA125, and only nine cases had CA125 values above the normal range. Without uniform nomenclature and diagnostic criteria, so many diagnostic names of this disease also cause confusion. The purpose of our study is to report a case of ACUM admitted to the First Affiliated Hospital of Guangxi Medical University and put forward our opinion on the nomenclature, diagnostic criteria, pathogenesis, and treatment of the disease.

2. Case Presentation

An 18-year-old adolescent female, gravida 0, was admitted to our hospital for excruciating dysmenorrhea since she was 17 years old. Menarche occurred when she was 14 years old, since then, she obtained a regular menstrual cycle (for 28 to 30 days) and a regular menstrual period (for 6 days) with normal menstrual flow. On March 18, 2020, she was diagnosed with the pelvic inflammatory disease at the local hospital. Nonsteroidal anti-inflammatory drugs (NSAIDs) were administered to relieve her symptoms, but her menstrual cramps did not improve. On March 4, 2021, she was diagnosed with focal adenomyosis based on the transabdominal ultrasound findings of a 35×29×32mm uterine cystic mass on the right side of the uterine body at our hospital, and gestrinone was administered for her in the next 3 months. Her symptoms subsided within 3 months of taking gestrinone but recurred after withdrawal.

On July 5, 2021, she was admitted to our hospital again for uncontrollable dysmenorrhea and called for a more definitive therapeutic regimen. Transrectal, three-dimensional, and transvaginal ultrasound detected an anechoic cystic lesion within a mixed echogenic mass located in the myometrium of the front right side of the uterine body, without connecting normal uterine cavity (Figure 1). The pelvic MRI was done to further characterize the adnexal mass. T1-weighted images revealed the cyst within the adnexal mass as an area of high signal intensity, while T2-weighted images showed it as an area of low signal intensity (Figure 2). She was diagnosed with ACUM based on the above clinical manifestations, as well as the results of ultrasound and MRI. After learning that surgery was the most effective solution to her problem, the patient requested surgery. After evaluation of the patient with indications for surgery and no contraindications, we performed conventional multi-incision laparoscopic surgery (MILS) on her on July 14, 2021.

Our procedure for MILS was performed as follows: (1) The patient was placed in the reverse Trendelenburg position under general anesthesia and endotracheal intubation; (2) A 1cm transverse incision was made subcutaneously along the upon margin of the umbilicus, where a pneumoperitoneum needle inserted into the abdominal cavity, and then 2.0 L of carbon dioxide was used to inflate the abdomen; (3) A 10mm Trocar was inserted through the primary incision, where a laparoscope was put into. The second, third, and fourth puncture points were made in the avascular zone of the hypogastrium; (4) Under the laparoscopic vision, an irregular uterine shape was observed, and a 20×20×30mm mass was observed at the junction of the right uterine round ligament and the right fallopian tube (Figure 3A). Bilateral ovaries and fallopian tubes were normal in appearance; (5) Six units of pituitrin diluted with normal saline were locally injected into the myometrium at the junction of the normal uterus and ACUM (Figure 3B). The serosa and myometrium at the most prominent point of the mass were incised using electrocautery to separate the mass (Figure 3C, D, E). Thereafter, the bottom myometrium of the mass cavity and the serosa were closed with a continuous suture using 2-0 barbed wire (Kehui) to restore uterine shape (Figure 3-F). The above puncture points were sutured using 3-0 monofilament absorbable thread. The woman's postoperative course was uneventful and she was discharged 2 days after surgery. Her dysmenorrhea had improved significantly.

Postoperative pathological results showed that the specimen was smooth tissue with endometrial glands and blood (Figure 4). The inner layer of the cavity was lined by endometrial glandular epithelial cells and stroma, and the outer layer appeared similar to normal myometrium. The patient was discharged on postoperative day 2 without complications. We learned that the dysmenorrhea significantly improved and the patient was followed up closely. Our follow-up indicated that the symptom was significantly relief without drug therapy after sixty days postoperatively.

3. Results

In 1996, Tamura et al.[10] proposed the concept of juvenile cystic adenomyoma (JCA) for the first time, pointing out that this disease is a rare disease that mostly occurs in nulligravid females and can cause dysmenorrhea. Since then, this kind of uterine cysts gradually arouses controversy among scholars all over the world. In 2010, Takeuchi et al. proposed the diagnostic criteria for JCA: First, the onset age ≤30 years; Second, cystic lesions ≥10mm in diameter that did not communicate with the normal uterine cavity; Third, associated with severe dysmenorrhea[11]. In the same year, by searching in the MEDLINE, Acién, et al.[12] suggested that most published cases of JCA, as well as those cases named noncommunicating accessory uterine cavities or uterine-like masses, are actually the same pathology: an accessory and cavitated uterine mass within an otherwise normal uterus, thus he terming it as ACUM. He proposed new criteria for ACUM in 2012: First, an isolated accessory mass containing a lumen; Second, uterus, tubes, and ovaries were normal; Third, surgical resection and pathological analysis of the lesion; Fourth, accessory cavity lined by endometrial epithelium with glands and stroma; Fifth, The content was chocolate-like liquid; Sixth, no adenomyosis (if uterus removed), but there could be small focus of adenomyosis in the myometrium adjacent to the accessory cavity[13]. The pathogenesis of ACUM is still controversial. At present, there are three main theories about pathogenesis,[14] including congenital anomaly theory, heterotopias theory, and metaplasia theory, but most scholars agree that ACUM is a congenital anomaly related to Mullerian duct[15].

We use the following search formula in PubMed: Accessory and Cavitated Uterine Mass OR ACUM OR JCA OR Juvenile cystic adenomyoma OR non-communicating accessory uterine cavities OR uterine-like masses OR cystic adenomyoma. Twenty-seven relevant studies including forty-five cases from 2000 to 2021 were obtained. After excluding four cases that didn't line with the ACUM diagnostic criteria proposed by Acién in 2012, forty-one cases of ACUM have been reported in the present literature. Among the four cases excluded by us, one case was excluded because her symptoms appeared after curettage of cornual pregnancy, and we considered that her symptoms mostly attributed to endometriosis caused by curettage instead of ACUM. Another three cases did not meet the second diagnostic criteria of ACUM proposed by Acién, two cases underwent oophorectomy due to previous endometriosis of the ovary, and one case was excluded because of intraoperative endometriosis of bilateral ovary and round ligament. Table 1 reveals the main clinical findings of the forty-one cases with ACUM that have been reported in previous literature. And we add a case of an 18-year-old woman (gravida 0, para 0) with over 1-years' history of progressive dysmenorrhea. In our case, we use the term ACUM and the diagnostic criteria proposed by Acién. After the minimally invasive laparoscopic resection of the lesion, dysmenorrhea significantly improved in our case.

Table 1
Review of literature related to ACUM

Author	Age at diagnosis (year)	Onset age (year)	GP	Symptom	CA125 (U/ml)	size of cyst(mm)	lesion size(mm)	imaging method	lesion position	diagnosis	medic therap
Nabeshima H[19]	19	NA	G0P0	severe dysmenorrhea	40.9	30	NA	TVU、MRI、HSG	in the right side of the uterus	Cystic adenomyoma	GnRHa
Takeda A[20]	20	13	G0P0	severe dysmenorrhea that began with menarche	25	15	30	TVU、MRI、HSG、DIP	in the right anterior portion of the uterine corpus caudal to the round ligament	Cystic adenomyoma	NSAID GnRHa
	20	14	G0P0	severe dysmenorrhea that began with menarche	40.5	11	26	TVU、MRI、HSG、DIP	in the left anterior portion of the uterine corpus caudal to the round ligament	Cystic adenomyoma	NSAID
Takeuchi H[11]	30	NA	G0P0	Pelvic pain	43	23	35	TVU, MRI	Three patients had the mass in the left side of the uterus and six patients on the right.	Cystic adenomyoma	N
	29	NA	G0P0	Pelvic pain and dysmenorrhea	141	15	30	TVU, MRI		Cystic adenomyoma	N
	27	NA	G2P2	Dyspareunia	36	19	42	TVU, MRI		Cystic adenomyoma	N
	20	NA	G0P0	Pelvic pain and dysmenorrhea	551	15	28	TVU, MRI		Cystic adenomyoma	OC
	30	NA	G2P2	Dyspareunia	34	15	30	TVU, MRI		Cystic adenomyoma	N
	28	NA	G0P0	Pelvic pain	12	19	25	TVU, MRI		Cystic adenomyoma	OC, Gr
	23	NA	G0P0	Pelvic pain	157	18	28	TVU, MRI		Cystic adenomyoma	OC
	20	NA	G0P0	Pelvic pain	34	10.5	40	TVU, MRI		Cystic adenomyoma	GnRHa
	20	NA	G0P0	Pelvic pain	14	16	34	TVU, MRI		Cystic adenomyoma	GnRHa
Akar ME[7]	15	15	G0P0	A 6-month history of intermittent episode right-sides periumbilical pain and severe dysmenorrhea	NA	25	47	CT, TSU,	in the right anterior fundal portion of the uterus	JCA	aroma inhibit proges OC, Gr
tijani[16]	35	35	G0P0	Intense pain in the left iliac fossa and pelvic	161	NA	21	USG	in the posterior fundus of the uterus and posterior face of the bladder	Giant uterus-like mass of the uterus	NA
Kriplani A[8]	16	14	G0P0	severe secondary dysmenorrhea	NA	NA	38	MRI, USG	In the right uterine wall near fundus	JCA	OC, NS mifepr
	18	17	G0P0	severe secondary dysmenorrhea	NA	NA	42	MRI, USG	In the right uterine wall near fundus	JCA	OC, NS
	16	15	G0P0	severe secondary dysmenorrhea	NA	NA	31	MRI, USG	In the anterior myometrium of the uterus	JCA	NSAID

Note: G=gravida; P=parity; N=no; NA=not available; USG=ultrasonography; CT=computer tomography; TAU=transabdominal ultrasound; TVU=transvaginal ult magnetic resonance imaging; HSG=hysterosalpingography; JCA= juvenile cystic adenomyoma; ACUM= accessory and cavitated uterine mass; OC=oral contr nonsteroidal anti-inflammatory drugs, IUD=intra uterine device.

Author	Age at diagnosis (year)	Onset age (year)	GP	Symptom	CA125 (U/ml)	size of cyst(mm)	lesion size(mm)	imaging method	lesion position	diagnosis	medic therap
	24	15	G0P0	severe secondary dysmenorrhea	NA	NA	30	MRI, USG	In the right uterine wall near fundus	JCA	OC, NS
Chun SS[21]	19	12	G0P0	pelvic pain and progressive dysmenorrhea	NA	21	30	MRI	In the left posterior uterine fundus	JCA	NSAID
Acién P[13]	36	19	G2P2	abdominal pain that was more intense in the left iliac fossae and increased during menstruation	NA	NA	50	TVU	in the left front side of the uterus	ACUM	OC
	18	14	G0P0	left iliac fossae pain, hypogastric pain and progressive dysmenorrhea	NA	NA	26	MRI, TRU,	on the left side of the uterine body	ACUM	NA
	19	10	G0P0	pelvic pain and progressive dysmenorrhea	NA	NA	20	TVU, HSG	in the anterior surface of the left horn	ACUM	OC, NS
	20	15	G0P0	iliac fossae pain and progressive dysmenorrhea	NA	3.5cm	40	TVU, MRI, HSG	In the anterior left side of the uterus	ACUM	analge drugs, NSAID
Branquinho MM[22]	17	15	G0P0	the right lower abdominal pain	NA	NA	3.3	TVU, TAU, MRI	within the right uterine myometrium and adjacent to the right ovary	JCA	analge OC
Jain N[23]	19	19	G0P0	severe dysmenorrhea and menorrhagia	NA	NA	NA	TAU	In the anterior right side of the uterus	cystic adenomyoma	OC, NS
Kumakiri J[4]	20	15	G0P0	severe dysmenorrhea	NA	30	NA	TVU, MRI	In the right side of the uterus	JCA	OC, NS
Bedaiwy MA[24]	16	NA	G0P0	severe dysmenorrhea and cyclic pelvic pain	NA	NA	30	USG, MRI	in the left anterior wall of the uterus	ACUM	OC, NS
Jain N[1]	24	NA	G0P0	severe dysmenorrhea and chronic pelvic pain	NA	NA	40	USG, MRI	in the right adnexa between the uterus and right ovary	ACUM	OC, NS
Pontrelli G[25]	27	NA	G0P0	menometrorrhagia, severe dysmenorrhea, dyspareunia, and chronic pelvic pain	96	80	NA	USG, MRI	In the posterior wall of the uterus	ACUM	Estrog and proges
Pabuccu EG[9]	20	20	G0P0	severe dysmenorrhea	NA	NA	NA	NA	in the right anterior wall just below the cornual end	JCA	NA
Garofalo A[26]	17	15	G0P0	severe pelvic pain and progressive dysmenorrhea	NA	17	28	USG, MRI	in the anterior right side of the uterus	ACUM	OC, NS

Note: G=gravida; P=parity; N=no; NA=not available; USG=ultrasonography; CT=computer tomography; TAU=transabdominal ultrasound; TVU=transvaginal ult magnetic resonance imaging; HSG=hysterosalpingography; JCA= juvenile cystic adenomyoma; ACUM= accessory and cavitated uterine mass; OC=oral contr nonsteroidal anti-inflammatory drugs, IUD=intra uterine device.

Author	Age at diagnosis (year)	Onset age (year)	GP	Symptom	CA125 (U/ml)	size of cyst(mm)	lesion size(mm)	imaging method	lesion position	diagnosis	medic therap
Dadhwal V[27]	23	20	G0P0	severe dysmenorrhea, severe episodic pain in the lower abdomen for 2 months and severe dysmenorrhea for 3 years.	NA	NA	39	TAU, MRI	in the anterior wall of the uterus near the cornual end	ACUM	NA
	16	13	G0P0	acute episodic pain in the left lower abdomen, the pain was severe in nature and occurred every 2-3 months for the 3 years	NA	NA	40	TAU, MRI	over the left uterine wall near the cornual end just below the insertion of the round ligament;	ACUM	NA
Strelec M[24, 28]	14	NA	G0P0	asymptom	NA	20	40	USG, MRI	NA	ACUM	N
Protopapas A[17]	14	13	G0P0	intolerable dysmenorrhea	NA	NA	38	MRI	In the left side of the uterus and extended to the uterine isthmus	JCA	OC
Wilcox A[29]	18	13	G0P0	severe dysmenorrhea	NA	NA	23	TVU, MRI	in the anterior left upper region of the uterus	JCA	OC, IU
	18	NA	G0P0	pelvic pain	NA	NA	36	TVU	within the left side of the uterus	JCA	OC
Kiyak H[30]	27	NA	G2P1	pelvic pain and delayed menses	NA	21	45	TVU	in the right cornual area	NA	NA
Minelli F[31]	19	18	G0P0	pelvic pain and dysmenorrhea	NA	NA	27	MRI, USG,	in the middle of the anterior wall of the uterus	JCA	Hormo therap
Iranpour P[32]	14	NA	G0P0	chronic recurrent pelvic pain	NA	NA	35	CT, TVU, MRI	In the left side of the uterus	ACUM	OC, NS
Arya S[6]	18	NA	G0P0	gradually worsening pelvic pain	NA	NA	30	CT, TVU, MRI	in the left lateral myometrium	JCA	Hormo therap
	16	NA	G0P0	worsening of chronic dysmenorrhea	NA	NA	47	CT, USG,	in the right side of the uterus	JCA	OC
Present study	18	17	G0P0	Severe dysmenorrhea				TAU, TRU, TVU, MRI	in the myometrium of the right side of the uterus	ACUM	Proges NSAID

Note: G=gravida; P=parity; N=no; NA=not available; USG=ultrasonography; CT=computer tomography; TAU=transabdominal ultrasound; TVU=transvaginal ult magnetic resonance imaging; HSG=hysterosalpingography; JCA= juvenile cystic adenomyoma; ACUM= accessory and cavitated uterine mass; OC=oral contr nonsteroidal anti-inflammatory drugs, IUD=intra uterine device.

We thought the term JCA and the diagnostic criteria of Takeuchi limit the age to females younger than 30 years old, but in fact, there were some reports on patients over 30 years old with ACUM[13, 16]. In addition, ACUM has a unique pathogenic site, which is located in the anterior lateral wall of the uterus near the corner of the uterus, and the size of the cyst within ACUM was all ≥0.5cm according to the current literature. Thus, we suggest that the pathogenic site of ACUM and the size of the cyst within ACUM should be included in the diagnostic criteria. What's more, we tend to agree that ACUM is a congenital disease since dysmenorrhea usually occurs within 5 years after menarche[3]. The patient in our case was initially diagnosed with focal adenomyosis, however, ACUM is easily misdiagnosed as obstructed cavitated rudimentary uterus horn in fact. Both of them present as a cavitated mass lined with endometrium, the within of which is hematometra. HSG can be done to distinguish the two. In the cases of non-communicating rudimentary horn, the fallopian tube of the affected side can't visualize on HSG[17].

It was considered that all congenital uterine anomalies have been implicated as a potential cause of infertility and adverse pregnancy events in a previous study[18]. Unfortunately, there is still a lack of literature that explore the reproductive outcomes of ACUM on patients. Since ACUM is a rare form of congenital uterine anomalies, the continuity of the myometrium and even the endometrium may have been disrupted. No matter how perfect the surgery is, there will be a scarred uterus after the surgery, which has a certain impact on the reproductive outcome of patients. But it still needs to be confirmed by long-term observational studies.

According to the existing literature, a total of forty-two cases of ACUM have been reported around the world, of which only fourteen cases provided a result of the patient's CA125, and nine cases had CA125 values above the normal range. Is the evaluation of CA125related to the occurrence of ACUM? We have no answer, because there are scarce reports of ACUM and even more scarce cases providing CA125. And it is unfortunate that our case didn't provide the information about CA125. More cases are needed to explain the relationship between ACUM and CA125.

4. Conclusion

ACUM is a rare form of uterine anomaly related to the malformation of Mullerian duct, which can cause severe dysmenorrhea and chronic pelvic pain. Surgical excision of the ACUM is still the most effective treatment, and laparoscopic surgery is proved to be a feasible and safe way. In our case, the patient benefits a lot after laparoscopic surgery. However, we can't answer whether the elevation of CA125 in some patients is related to ACUM and whether it has a certain impact on the reproductive outcome of patients now. It calls for more cases and research to deal with these problems. Anyways, our report adds a case to the current literature, and we also tend to agree that ACUM is a congenital disease since dysmenorrhea occurs within 5 years after menarche in our patient. In addition, we put forward that the pathogenic site of ACUM and the size of the cyst within ACUM should be included in the diagnostic criteria.

Declarations

Authors' contributions

H.H wrote the first draft of the manuscript; X.L and H.H provided figures; X.L and J.F designed research, critically revised the paper, and provided funding support. All Authors read and approved the final version of the manuscript

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Availability of data and materials

The data that support the findings of this article are available from the corresponding author upon reasonable request.

Ethics approval and consent to participate

Consent for the use of anonymized data and imaging was obtained from the next of kin.

Consent for publication

All authors read and approved the final manuscript.

Competing interests

Written informed consent was obtained from the patient for publication of this case report any accompanying images. The authors declare no competing interests.

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Figures

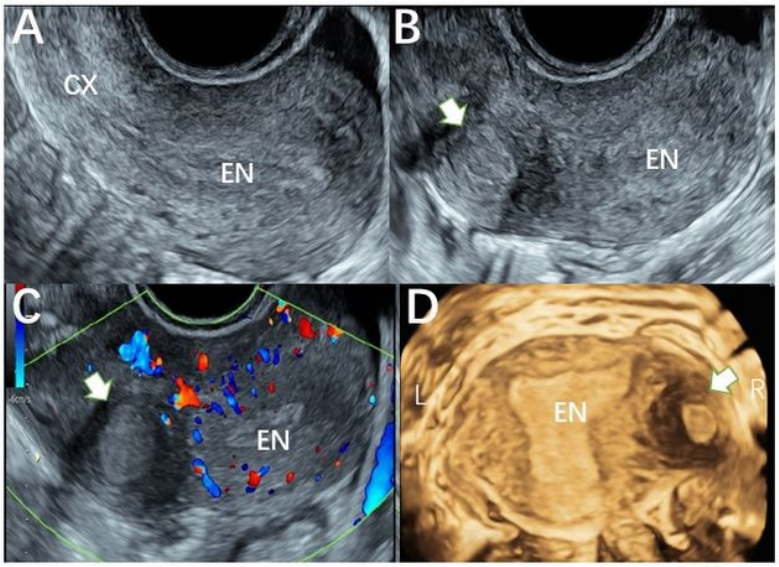

Figure 1

(A) Transvaginal ultrasound in a longitudinal section, (B) Transvaginal ultrasound in a transverse section, (C) Transrectal ultrasound and (D) Three-dimensional ultrasound show the ACUM (white arrow) located in the myometrium of the right side of the uterus. (CX: Cervix, EN: Endometrium, L: Left, R: Right.)

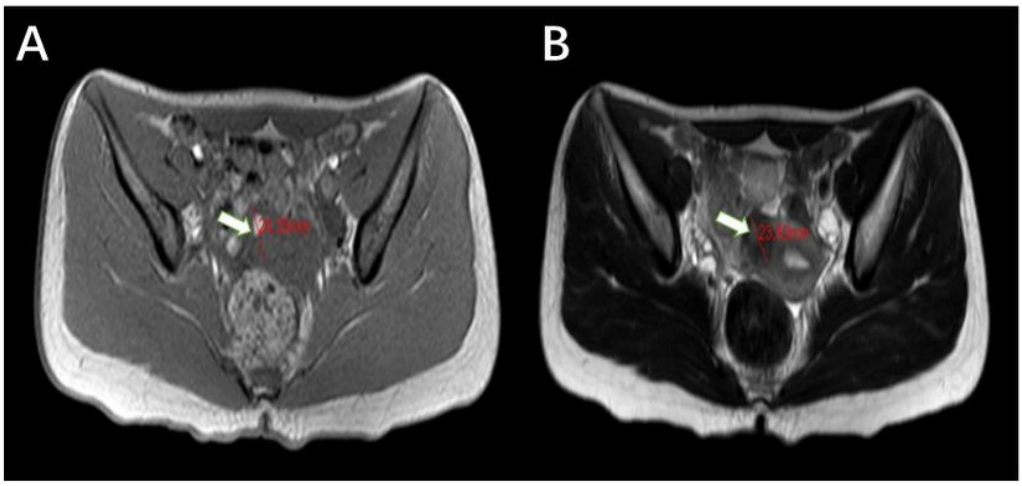

Figure 2

(A)T1-weighted MRI of the content within ACUM (white arrow) was high signal intensity. (B)T2-weighted MRI of ACUM (white arrow) was low signal intensity.

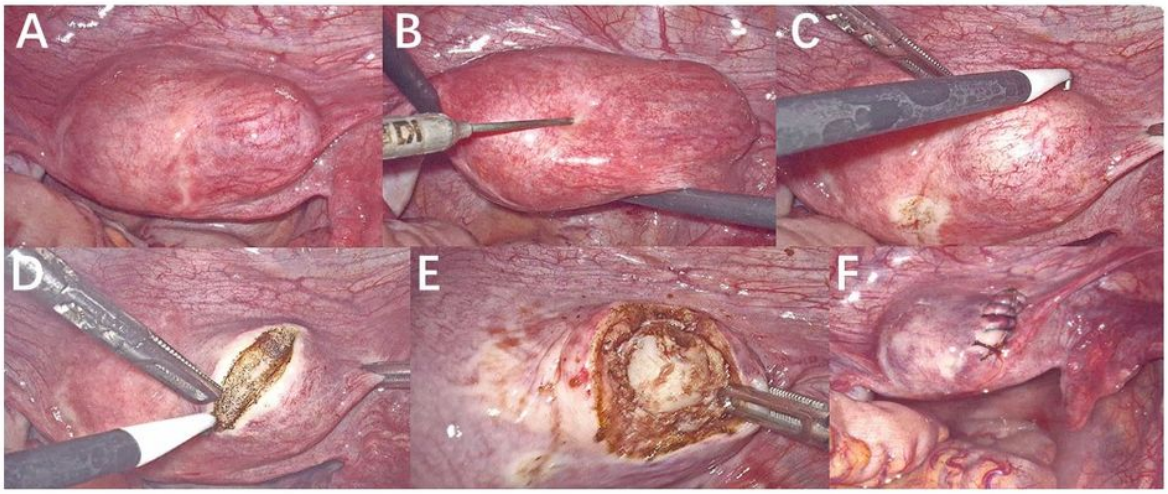

Figure 3

(A) A 2*2*3cm mass was observed at the junction of the right uterine round ligament and the right fallopian tube. (B) Six units of pituitrin diluted with normal saline were locally injected into the myometrium. (C, D, E) An incision between the normal myometrium and lesion was performed. (F) The surface of the wound in the uterine serosa sutured using unidirectional barbed string.

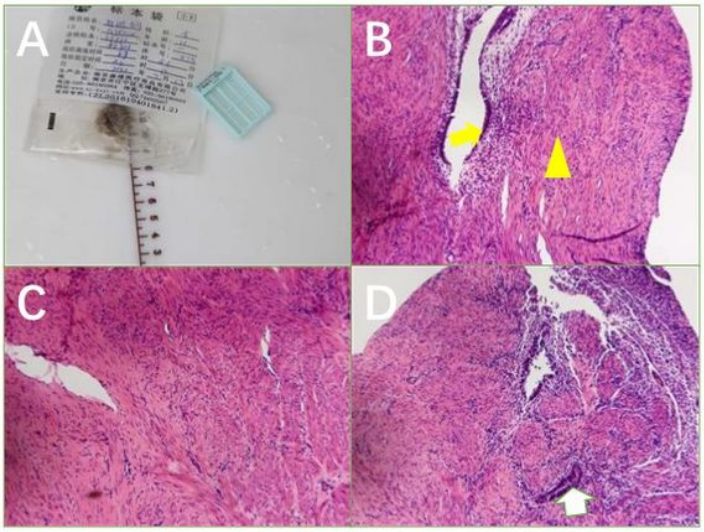

Figure 4

(A) Excised lesions. (B) The inner layer was lined by endometrial glandular epithelial cells and stroma (yellow arrow), and the outer layer appeared similar to normal myometrium (yellow triangle). (C) The myometrium is irregularly arranged around the cyst. (D) Myometrium with endometrial gland invasion (white arrow).