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# Submucosal cystic adenomyosis: a report of five cases and review of the literature

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## Abstract

**Aim** To investigate the clinical characteristics, diagnosis, and clinical treatment of submucosal cystic adenomyosis.

**Methods** The clinical data of five cases of patients with submucosal cystic adenomyosis in our hospital from January 2020 to June 2023 were retrospectively analyzed.

**Results** The average age of the patients was 37.8 ± 4.5 years old, three of them experienced prolonged menstruation and heavy menstrual bleeding. All patients had a history of abnormal uterine bleeding and mild to moderate dysmenorrhea, with a VAS score of 2.8 ± 1.6. The average Carbohydrate antigen 125 (CA125) value was 29.9 ± 23.6U/ml. Two out of the five patients (40%) had CA125 values above the upper limit of normal. The nodules had a diameter of 3.2 ± 1.3 cm and a cavity size of 1.3 ± 0.7 cm. Color ultrasound revealed hypo or iso or anechoic echoic cysts, and blood flow signals were detected. The magnetic resonance imaging (MRI) findings varied among each patient. All the patients underwent hysteroscopy and resection of uterine cavity-occupying lesions, and no recurrence was observed.

**Conclusions** The clinical features of submucosal cystic adenomyosis include abnormal uterine bleeding and menstrual changes, and the degree of dysmenorrhea is generally not severe. The diagnostic utility of CA125 in submucosal cystic adenomyosis may be limited. The three-dimensional ultrasound and MRI are valuable preoperative examination methods currently. Hysteroscopy can not only diagnose submucosal cystic adenomyosis, but also treat it, and preserve the fertility function of the patient.

**Keywords** Submucosal cystic adenomyosis, Adenomyosis, Abnormal uterine bleeding, Dysmenorrhea, Hysteroscopy

## Background

Adenomyosis is a common disease in gynecology characterized by the diffuse or localized invasion of endometrial glands and stroma into the myometrium. Extensive research indicates that adenomyosis has been observed in a range of 8.8% to 61.5% of hysterectomy specimens over the past five decades [1]. Cystic adenomyosis, a rarer form of adenomyosis, is distinguished by the presence of one or more cystic cavities within the myometrium. These cavities contain brownish, stale bloody fluid and do not connect with the uterine cavity. The cyst wall is composed of endometrial glands and epithelium of the mesenchyme [2]. The main clinical manifestations of female cystic adenomyosis are dysmenorrhea and chronic pelvic pain, which negatively affect their quality of life. But so far, its etiology and pathogenesis are not clear [3]. Even the literature on the analysis and summary of its clinical characteristics is rare. Because of the low incidence of the disease, the literature on cystic adenomyosis is confined

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to case reports and several small series. Thus, the rates of misdiagnosis and missed diagnosis are often high. Most of the case reports from recent years revealed that the lesions are located in the uterine muscular wall or subserosa [4]. Submucosal cystic adenomyosis, as a special subtype of cystic adenomyosis, does not have much literature about it at present. Since Dobashi, Y [5] first reported polypoid cystic adenomyosis in 1992, less than five cases of submucosal cystic adenomyosis have been reviewed [6]. However, these case studies do not provide a comprehensive overview of the clinical characteristics of submucosal cystic adenomyosis, particularly in relation to the efficacy and projected outcomes of hysteroscopic treatment [7]. In this paper, we studied the cases of submucosal cystic adenomyosis treated in our hospital, aiming to explore the clinical characteristics of the disease and the effect of hysteroscopic surgery. Our findings furnish preliminary clinical evidence that warrants further exploration into submucosal cystic adenomyosis in future research.

## Methods

From January 2020 to June 2023, five patients with pathologically diagnosed submucosal cystic adenomyosis were admitted to Fujian Maternity and Child Health Hospital. The age of onset, disease duration, age of menarche, clinical manifestations (dysmenorrhea and menstrual condition changes), fertility history, family history, prior drug treatment, preoperative uterine cavity surgery history, Carbohydrate antigen 125 (CA125) value, results of the three-dimensional color ultrasound examination, magnetic resonance imaging (MRI) examination results, hysteroscopic surgery data (nodule number, nodule position, and percentage of lesions that indented the endometrial cavity), and follow-up results of all patients were collected.

The visual analog scale (VAS) was used to score patients' dysmenorrhea pain. 0 was classified as no pain, 1–4 as mild pain, 5–6 as moderate pain, 7–9 as severe pain, and 10 as extreme pain.

The procedures of hysteroscopic surgery were as follows: the patient was given general anesthesia, placed in the lithotomy position, the dilation fluid used was normal saline, the dilation pressure was set at 20kpa, and the uterine mass was completely removed using a ring electrode under hysteroscopy.

The follow-up was mainly conducted by combining regular outpatient reviews and telephone follow-ups. The start time of the follow-up was the operation time, and the end time of the follow-up was December 2023. The follow-up included clinical manifestations (such as dysmenorrhea and menstruation condition changes), recurrence (based on gynecological examination and transvaginal ultrasonography), CA125 value, pregnancy status, etc. *SPSS* 23.0 statistical software was used for data processing and analysis.

## Results

### General information

From January 2020 to June 2023, a total of five cases of submucosal cystic adenomyosis were admitted to the gynecological ward of our hospital (Table 1 shows general characteristics). The average age of the patients was 37.8 ± 4.5 years old, the onset age was 37.4 ± 4.7 years old, and the disease duration was 3.0 ± 2.3 months.

All five patients had menarche when they were 14–15 years old, and three of them experienced prolonged menstruation and heavy menstrual bleeding. Additionally, all patients reported mild to moderate dysmenorrhea. The visual analog scale (VAS) was used to measure pain, and the average pain score was 2.8 ± 1.6. One patient had a maximum score of 6. Furthermore, all patients had a history of abnormal uterine bleeding that did not respond to medical treatment. All of the patients were married women with children and had a history of uterine procedures, such as curettage due to embryo termination or placental retention, and hysteroscopic resection of submucosal myoma. Three patients had a positive familial history of adenomyosis, and each patient had previously used condoms to prevent pregnancy. Each patient's previous history of pregnancy, previous methods of preventing pregnancy and premature abortion, family history of adenomyosis and uterine cavity surgery is shown in Table 2.

### Auxiliary examinations

Preoperative color ultrasonography showed that the nodules were 3.2 ± 1.3 cm in diameter and 1.3 ± 0.7 cm in cavity. The three-dimensional color ultrasound is characterized by hypo, iso, or anechoic cysts, and can detect blood flow signals (Fig. 1). However, the MRI findings of each patient were not exactly the same. Case 1 had a cystic lesion that was hyperintense on T1-weighted and T2-weighted (Fig. 2). Case 2 did not have an MRI. Case 3 had a mixed signal cystic lesion, partly hypointense on T1-weighted and hyperintense on T2-weighted, and partly hyperintense on T1-weighted and hyperintense on T2-weighted. Case 4 had a cystic lesion with isointensity on T1 and slightly hyperintensity on T2. Case 5 had a nodule that was hypointense on T1-weighted and hyperintense on T2-weighted. The average CA125 value was 29.9 ± 23.6U/ml. Two out of the five patients (40%) had CA125 values above the upper limit of normal (> 35U/ml),

**Table 1** General characteristics

| Case | Age(years) | Age of onset (years) | Disease duration (months) | Menarche (years) | Dysmenorrhea | VAS Pain Score | Heavy menstrual bleeding | Prolonged menstruation | Menstrual cycle length(day) | Menstrual period days | Abnormalvaginal bleeding | Prior drug treatment |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 44 | 44 | 2 | 15 | Exist | 2 | Exist | Exist | 30 | 8 | Exist | Tranexamic acid |
| 2 | 37 | 37 | 1 | 14 | Exist | 1 | Null | Null | 28 | 7 | Exist | None |
| 3 | 32 | 31 | 6 | 14 | Exist | 4 | Exist | Exist | 30 | 10–12 | Exist | Dydrogesteron,Drospirenone and ethinylestradiol tablets (II) |
| 4 | 40 | 39 | 5 | 14 | Exist | 5 | Exist | Exist | 30–40 | 7 | Exist | Dydrogesteron |
| 5 | 36 | 36 | 1 | 14 | Exist | 2 | Null | Null | 30–38 | 7–8 | Exist | None |
| mean ± SD or Median[a] | 37.8 ± 4.5 | 37.4 ± 4.7 | 3.0 ± 2.3 | 14(14,14) | | 2.8 ± 1.6 | | | | | | |

[a] Data are shown as mean ± standard deviation for normal distributions, or median with interquartile range for non-normal distributions

**Table 2** History of pregnancy, previous methods of preventing pregnancy and premature abortion, family history of adenomyosis and uterine cavity surgery

| Case | Gestational | Produce | Mode of delivery | History of pregnancy loss | Previous methods of preventing pregnancy and premature abortion | Family history of adenomyosis | History of uterine cavity surgery |
|---|---|---|---|---|---|---|---|
| 1 | 5 | 3 | Vaginal delivery | 2 | Male condoms | Exist | Hysteroscopy, Fractional curettage of uterus |
| 2 | 3 | 1 | Caesarean delivery | 2 | Male condoms | Null | Hysteroscopic myomectomy, Curettage due to embryo cessation |
| 3 | 1 | 1 | Vagina delivery | 0 | Male condoms | Exist | Hysteroscopic myomectomy |
| 4 | 2 | 2 | Vaginal delivery | 0 | Male condoms | Exist | Hysteroscopic myomectomy, Laparoscopic adenomyomectomy |
| 5 | 2 | 1 | Vaginal delivery | 1 | Male condoms | Null | Laparotomic hysteromyomectomy, Hysteroscopy, Curettage due to placenta residue |

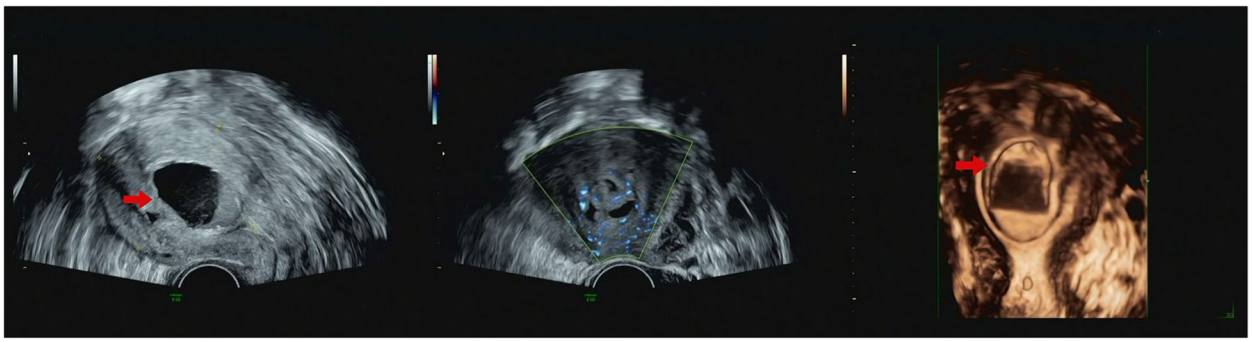

**Fig. 1** The three-dimensional color ultrasound examination in case1 reveals that the uterine nodule exhibited characteristics of inhomogeneous echogenicity, with hypo or anechoic properties, and the presence of streaks indicating blood flow signal surrounding the mass

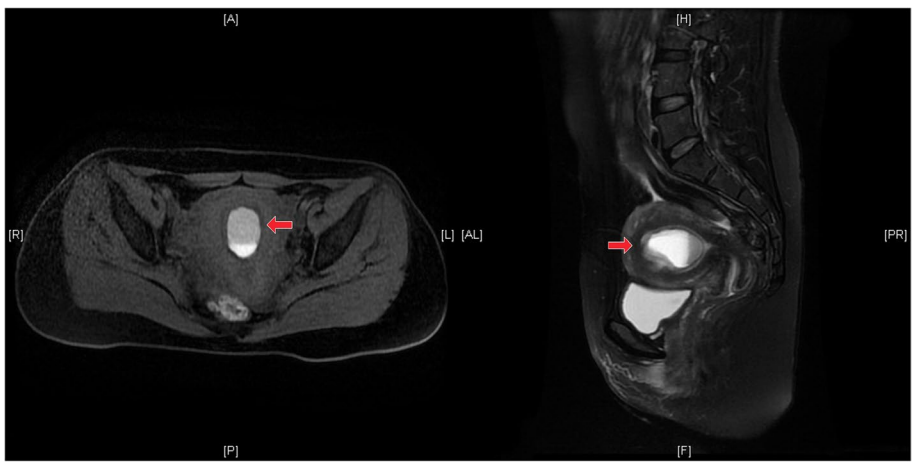


**Fig. 2** The MRI findings of Case1 reveal that the uterine mass exhibited characteristics of a cystic lesion, displaying hyperintensity on both T1-weighted and T2-weighted images. Liquid–liquid stratification can be observed in the capsule cavity

**Table 3** The characteristics of auxiliary examinations

| Case | CA125 value (U/ml) | Nodule diameter(cm) | Cystic cavity diameter(cm) | Ultrasound of blood flow | The three-dimensional color ultrasound manifestation | The location of the nodules shown on the three-dimensional color ultrasound | MRI features |
|---|---|---|---|---|---|---|---|
| 1 | 21.9 | 3.4 | 1.5 | Exist | Inhomogeneous echogenicity, hypo or anechoic | Anterior wall of the uterus | Cystic lesion hyperintense on T1-weighted and T2-weighted. |
| 2 | 9.3 | 2.5 | 1.7 | Exist | Anechoic | The bottom of the uterus | None |
| 3 | 63.1 | 4.0 | 1.3 | Rich | Inhomogeneous echogenicity, hypo or isoechoic | Uterine posterior wall | Mixed signal cystic lesion, partly hypointense on T1-weighted and hyperintense on T2-weighted, partly hyperintense on T1-weighted and hyperintense on T2-weighted. |
| 4 | 45.5 | 4.6 | 2.0 | Exist | Hypoechogenic | Left posterior wall of uterus | Cystic lesion, isointensity on T1 and slightly hyperintensity on T2. |
| 5 | 9.9 | 1.3 | 0.2 | Exist | Hypoechogenic | Uterine posterior wall | Hypointense on T1-weighted and hyperintense on T2-weighted. |
| mean ± SD | 29.9 ± 23.6 | 3.2 ± 1.3 | 1.3 ± 0.7 | | | | |

while three (60%) were within the normal range. The characteristics of auxiliary examinations are shown in Table 3.

### Surgical treatment

All patients underwent hysteroscopic resection of the uterine lesion. Intraoperative hysteroscopy showed only one lesion in each case, with two located in the posterior wall of the uterus, one in the anterior wall near the uterine floor, one in the uterine posterior wall near the bottom of the uterus, and one at the bottom of the uterus. The ultrasonographic determination of the lesion's location was completely consistent with what was observed during hysteroscopy, demonstrating an accuracy of 100%. The percentage of lesions that indented the endometrial cavity is 30–80% (The surgical characteristics are shown in Table 4). During the operation, after the uterine cysts were cut by the electrotomy ring, the chocolate-like fluid could be seen flowing out of the rupture of the cyst wall, and the inner wall of the cyst was covered by the endometrial tissue (Fig. 3). After the surgery, all patients were pathologically diagnosed with cystic adenomyosis (Figs. 4 and 5).

### Follow-up

All patients received 3 cycles of Gonadotropin-releasing hormone agonist (GnRH-a) postoperatively, and 3 patients had no fertility requirements and continued to use Levonorgestrel intrauterine system (Mirena). Postoperative dysmenorrhea symptoms were significantly

**Table 4** The surgical characteristics

| Case | Nodule number | Nodule position | Percentage of lesions that indented the endometrial cavity (%) |
|---|---|---|---|
| 1 | 1 | Anterior wall of the uterus near the bottom of the uterus | 80 |
| 2 | 1 | The bottom of the uterus | 30 |
| 3 | 1 | Uterine posterior wall near the bottom of the uterus | 50 |
| 4 | 1 | Uterine posterior wall | 80 |
| 5 | 1 | Uterine posterior wall | 60 |

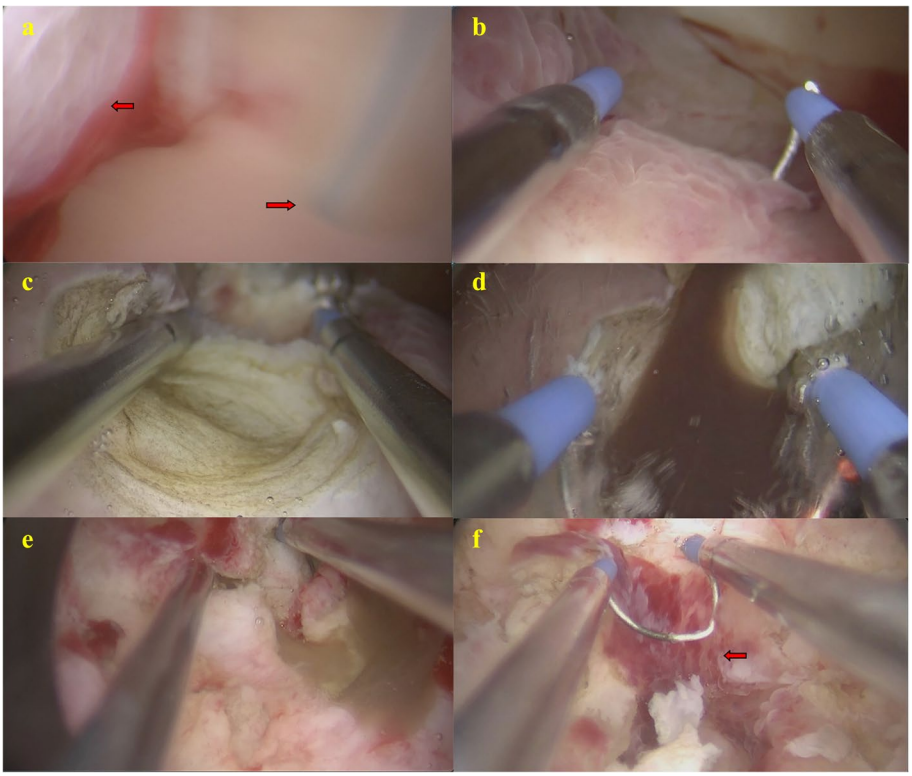


**Fig. 3** **a** During hysteroscopic surgery, a giant cystic mass is observed protruding into the uterine lumen on the anterior wall of the uterus. Arrows are used to indicate the nodule and the opening of the left fallopian tube. At this time, the left tubal hydrotubation is being performed with the use of methylene blue. **b**,**c** Submucosal cystic adenomyosis is progressively resected with an electrotomy ring under hysteroscope. **d**,**e** During the operation, after the uterine cysts are cut by the electrotomy ring, the chocolate-like fluid can be seen flowing out of the rupture of the cyst wall. **f** The inner wall of the cyst is covered by the endometrial tissue

improved in all patients, and the VAS score was 0–1. Additionally, abnormal uterine bleeding, heavy menstrual bleeding, and prolonged menstruation were also alleviated to varying degrees. The average CA125 value was 14.1 ± 7.0 U/ml. Two of the patients had fertility requirements and were trying to become pregnant, but were still not successful. The follow-up period ranged from 7 to 42 months until December 2023. All patients showed no recurrence and achieved a satisfactory curative effect. (Postoperative follow-up for each case is shown in Table 5).

## Discussion

Cystic adenomyosis is a special type of adenomyosis. Paruleka first reported the condition in 1990 [8]. Currently, there are only 100 cases reported in the worldwide literature. Recognition and awareness of this disorder are extremely limited, and even less information is available concerning submucosal cystic adenomyosis [6]. So far, the literature on submucosal cystic adenomyosis is confined to isolated case reports [6], with no comprehensive review available to collate the clinical features of this condition and the effectiveness of hysteroscopic treatment. We summarized the features of submucosal cystic adenomyosis and reviewed relevant literature.

### Definition, classification and pathogenesis of diseases

The pathogenesis of cystic adenomyosis remains poorly understood, and there is no uniform classification criterion. Brosens et al [9] classified cystic adenomyosis into three subtypes based on cyst location in the uterus: Type A (A1: submucosal/intermuscular, A2: polypoid), Type B (B1: subserous, B2: exogenous lesions), and Type C (uteroid mass).In this paper, preoperative three-dimensional colour doppler ultrasonography showed that the nodules protruded into the uterine cavity, and even the endometrium was compressed. According to the findings of hysteroscopy, submucosal cystic adenomyosis of the uterus could be diagnosed in all five patients.

In addition, most scholars tend to divide cystic adenomyosis into two types: adult type and adolescent type, based on different age groups. Congenital cystic adenomyosis primarily occurs in adolescents, with disease onset typically happening more than 5 years after

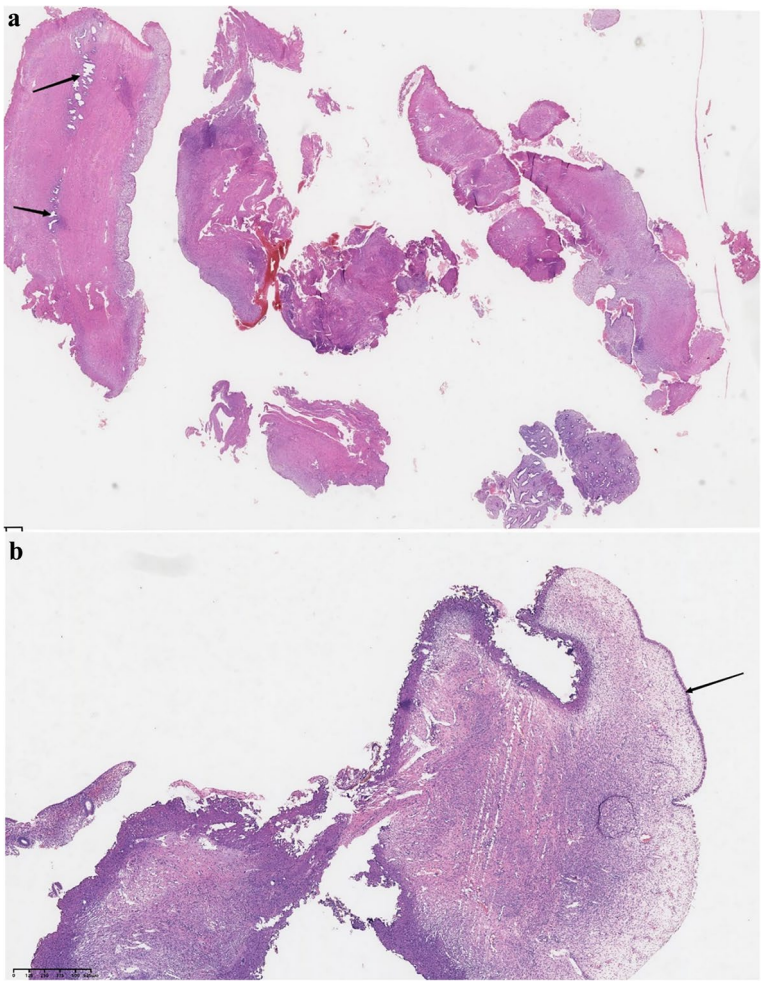


**Fig. 4** **a** In case 4's pathology section, arrows point to glands in the uterus's smooth muscle cells. **b** Endometrial cells lining the capsule wall are indicated by the arrow

menarche or before the age of 18 [10]. It may be the residual Mullerian duct in the myometrium under the action of estrogen that causes periodic bleeding to form a sac, resulting in the progressive aggravation of dysmenorrhea [11]. Kriplani et al. [12] proposed that compared with juvenile cystic adenomyosis, adult type patients were generally older than 30 years old, and most of them had a history of uterine operation and fertility, which is a high-risk factor for endometrial adhesion zone injury. Once the endometrial binding zone is damaged, the basal endometrium may invade the myometrium and eventually develop cystic adenomyosis [13]. In this study, the mean age of five patients diagnosed with submucosal cystic adenomyosis was 37.8 ± 4.5 years. The age of onset was 37.4 ± 4.7 years, indicating that all cases were of the adult type. Additionally, all patients had a history of childbirth and uterine manipulation. Two of them developed cystic adenomyosis in a nearby location after the resection of submucosal myoma from the posterior wall of the uterus, which appears to be consistent with the mechanism of acquired disease.

### Clinical features

The clinical presentations of submucosal cystic adenomyosis closely resemble those of classical adenomyosis, characterized by progressive dysmenorrhea, heightened menstrual volume, abnormal uterine bleeding, and chronic pelvic pain. However, some patients lack typical clinical presentation [13]. This may be related to the different location of the lesion and the diameter of the cyst.

Most of the dysmenorrhea in our patients was mild or moderate, and the dysmenorrhea in women with uterine cystic adenomyoma can be explained by the progressive increase in cyst size caused by repeated intracystic bleeding during menstruation. In patients with adenomyosis, FMT (fibroblast-to-myofibroblast transdifferentiation)

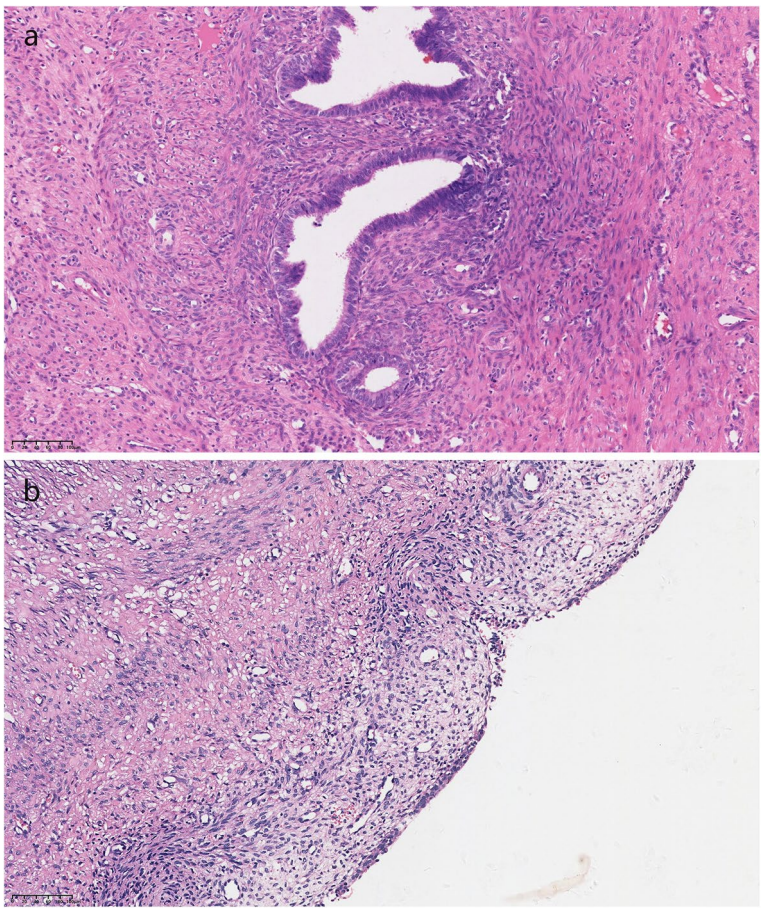


**Fig. 5** **a** and **b** are enlarged portions of Fig. 4a and b respectively

and SMM (smooth muscle metaplasia) occur in the uterus. These processes involve the transformation of ectopic endometrial stromal cells into smooth muscle cells, which then integrate into the muscle layer. This integration is likely to lead to irregular uterine contractions, causing spasmodic contractions and resulting in dysmenorrhea [14]. However, the focus of submucosal cystic adenomyosis is close to the endometrial protrusion towards the uterine cavity. Therefore, its impact on uterine contraction is not as significant as that of typical adenomyosis patients. As a result, the degree of dysmenorrhea is mostly relatively mild. Yan-Yan Fan's report on dysmenorrhea symptoms in intrauterine cystic adenomyosis indicates a mild to moderate severity [6], whereas the dysmenorrhea symptoms associated with subserous or muscular cystic adenomyosis are markedly more severe [15]. Additionally, this may be attributed to the fact that most of the patients in this study sought treatment promptly and did not have a long history of the disease.

All patients included in this study have a history of abnormal uterine bleeding, and three out of the five patients (60%) experienced increased menstrual volume and prolonged menstrual periods, which is consistent with previously reported findings [6]. These symptoms may be attributed to the following reasons: ① Compared with lesions close to the serous layer of the uterus, if the adenomyosis lesions are closer to the endometrium, the increase in endometrial surface area is more obvious, and fibrosis is more likely to involve the endometrium. This is more likely to lead to low endometrial repair function and prolong the endometrial repair time [16]. ②The larger intermuscular mass obstructs the closure of the broken blood vessels on the exfoliated surface of the endometrium due to its occupying effect, resulting in poor vasoconstriction [17]. ③Endometrial and venous muscle structure changes can lead to a disruption in venous plexus blood flow, resulting in endometrial hypercongestion [17]. However, these changes are also the reason why uterine submucous myoma is more likely to cause abnormal uterine bleeding and heavy menstrual bleeding, so it is important to be cautious and discerning.

**Table 5** Postoperative follow-up

| Case | Follow-up time (month) | Drug therapy in the postoperative period | VAS Pain Score | Heavy menstrual bleeding | Prolonged menstruation | Menstrual cycle length (day) | Menstrual period days | Abnormal vaginal bleeding | Pregnancy after surgery | Recrudescence | CA125 value (U/ml) |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 42 | GnRH-a 3 cycles + LNG –IUS | 0 | Null | Null | 30 | 5–6 | Null | No fertility requirement | No | 18.8 |
| 2 | 19 | GnRH-a 3 cycles | 0 | Null | Null | 28 | 3 | Null | Trying to get pregnant and still not pregnant | No | 7.2 |
| 3 | 19 | GnRH-a 3 cycles + LNG –IUS | 1 | Null | Null | 30 | 9–10 | Null | No fertility requirements | No | 23.7 |
| 4 | 15 | GnRH-a 3 cycles + LNG –IUS | 1 | Null | Null | / | / | Null | No fertility requirement | No | 12.4 |
| 5 | 7 | GnRH-a 3 cycles | 0 | Null | Null | 30 | 7 | Null | Trying to get pregnant and still not pregnant | No | 8.6 |
| mean ± *SD* or Median[a] | | | 0(0,1) | | | | | | | | 14.1 ± 7.0 |

[a] Data are shown as mean ± standard deviation for normal distributions, or median with interquartile range for non-normal distributions

### Auxiliary examinations and diagnosis

The auxiliary examination of submucosal cystic adenomyosis mainly includes imaging examination, pathological examination, and determination of blood-related tumor markers. Transvaginal three-dimensional ultrasound is highly valuable in the diagnosis of uterine diseases and various types of adenomyosis [18, 19]. Performing a transvaginal color Doppler ultrasound for the screening of intrauterine cystic adenomyosis is cost-effective and convenient. In ultrasound images, mixed masses and dark cystic fluid areas are visible [10]. Figure 1 is a series of typical three-dimensional color ultrasound images of submucosal cystic adenomyosis in the anterior wall of the uterus. In comparison to traditional transvaginal ultrasound, this technique offers enhanced color representation, more realistic stereoscopic images, and improved resolution. Consequently, it effectively eliminates the majority of blind spots during examinations and provides clinicians with a more intuitive three-dimensional reconstruction image, facilitating their comprehension of uterine lesions. As a result, clinicians can gain a better understanding of the anatomical correlation between uterine lesions and the endometrium and muscle layer. In this study, the transvaginal three-dimensional color ultrasound was used in all cases. The results indicated that the lesions were hypo, iso, or anechoic cysts with cystic cavities, detectable masses, and blood flow signals. It is largely consistent with the ultrasonic characteristics documented in prior literature [6, 20]. Additionally, the location of the lesions was also very accurate.

In terms of assisted diagnosis, MRI may be superior to ultrasound. The fluid component of the cyst is highly signaled on T1-weighted images, medium–high on T2-weighted images, and the surrounding hypertrophic muscle layer is low signaled on T2-weighted images [21]. Fugure2 is a typical MRI image of cystic adenomyosis. The intrauterine cysts show T1 and T2 hypersignal shadows. Liquid–liquid stratification can be observed in the capsule cavity. It has been suggested that this can also be distinguished from uterine myomatosis, which MRI showed a high signal in T1-weighted images and a low signal in T2-weighted images [22]. However, the MRI characteristics of each case in this study are not exactly the same, which may be related to the different duration of the disease and the formation time of sac fluid. Nonetheless, MRI still objectively reflects the position of the nodule and its relationship with the endometrium and myometrium, making it a key tool for identifying adenomyosis subtypes [23]. In addition to submucous myomatosis or adenomyoma of the uterus, the possibility of submucosal cystic adenomyosis should be considered when MRI suggests an intrauterine cystic cavity with mixed signals.

Similar to common adenomyosis, some patients with submucosal cystic adenomyosis also have elevated serum CA125 levels. According to certain literature, the presence of elevated pre-surgical CA125 levels and a thickened posterior myometrial layer were identified as independent risk factors for the manifestation of dysmenorrhea [24]. Certain scholars propose that there exists a positive correlation between the CA125 level and the level of dysmenorrhea [25]. Studies have indicated that the specificity and sensitivity of serum CA-125 levels in diagnosing submucosal cystic adenomyosis are limited [6]. However, elevated CA-125 levels can aid in distinguishing this condition from endometrial polyps and uterine myomatosis [26]. In this study, not all patients had elevated CA125 values, which is consistent with the fact that not all patients had severe dysmenorrhea. The presence of dysmenorrhea was notably observed in Case3 and Case4, thereby indicating a significant increase in CA125 levels.

Diagnosing submucosal cystic adenomyosis, like common cystic adenomyosis, involves both intraoperative observations and pathological analysis. Hysteroscopy is an important method for the diagnosis and treatment of uterine diseases. The histopathological features of submucosal cystic adenomyosis are: the skin of the capsule wall is arranged by the endometrial glands and the stroma, and the proliferative muscle tissue is covered around the capsule wall [7]. Figures 4 and 5 show a typical cystic adenomyosis pathology image.

### Treatment and prognosis

The therapeutic principle for submucosal cystic adenomyosis is the radical resection of the lesions, promoting fertility, and preventing recurrence. Treatment can vary depending on the age at onset, fertility request, lesion location, size, and symptoms [27]. For young patients or those with fertility requirements, focal removal is the preferred treatment. For the treatment of intrauterine cystic adenomyosis, hysteroscopic resection is preferred [28]. The boundaries of submucosal cystic adenomyosis are more clearly defined than those of typical adenomyosis, making complete resection of the lesion easier [29].

The lesion located under the superficial mucosa can be directly observed during hysteroscopy. The purplish-blue cyst protruding into the uterine cavity, seen under hysteroscopy, is a characteristic presentation of submucosal cystic adenomyosis [30]. Abnormal angiogenesis or fibrosis can be seen in the endometrium below the site of the cyst [7]. Hysteroscopic identification of intrauterine cystic adenomyosis is specific and can be distinguished from myomatous cystic degeneration and congenital uterine cysts. The focus of submucosal cystic adenomyosis is characterized by a chocolate-like fluid in the

cystic cavity [31]. In our study, we directly observed the outflow of old blood resembling chocolate during hysteroscopic electrotomy of the submucosal cystic adenomyosis lesion. Subsequently, we were able to observe the smooth, thick-walled cyst cavity (Fig. 3). There is an advantage to the hysteroscopic approach in that the outer myometrium is intact, and there is no abdominal scarring to be dealt with. Previous literature suggests that when smaller cystic structures are present in the inner third of the intramural part or submucosally, a minimally invasive hysteroscopy is recommended [7].

Current drug therapy for submucosal cystic adenomyosis mainly consists of analgesics and menstrual control. Nonsteroidal anti-inflammatory drugs, oral contraceptives, and gonadotropin-releasing hormone agonists (GnRH-a) only offer temporary relief and can lead to recurrence once the drug is discontinued. Additionally, common painkillers or hemostatic drugs are less effective in managing the condition [32].

None of the patients in this study had abnormal uterine bleeding after hysteroscopic complete resection of the lesion. Additionally, dysmenorrhea, increased menstrual volume, and prolonged menstrual cycles were greatly improved. The CA125 levels of the patients exhibited a notable decrease following the surgical procedure. Therefore, in our study, hysteroscopic resection of submucosal cystic adenomyosis can achieve a good therapeutic effect.

All patients received follow-up GnRH-a treatment after surgery, and three of them, who did not currently have fertility requirements, continued using Mirena after three cycles of GnRH-a. None of the five patients experienced a relapse. Two of the patients had fertility requirements and were preparing for pregnancy but were not yet pregnant, and further follow-up was needed for subsequent pregnancies. Several cases of successful treatment of submucosal cystic adenomyosis via hysteroscopy have been documented [6, 7]; however, there have been no reported instances of successful pregnancy following the surgical intervention.

Due to the small number of cases, the views proposed in this paper have certain limitations, and a larger sample size and clinical observational studies are needed to provide more comprehensive and precise clinical recommendations.

## Conclusions

Submucosal cystic adenomyosis is a relatively rare form of adenomyosis. The clinical features include abnormal uterine bleeding, menstrual changes, and varying degrees of dysmenorrhea. The diagnostic utility of CA125 in submucosal cystic adenomyosis may be limited. The three-dimensional color ultrasound and MRI are currently valuable preoperative examination methods. Hysteroscopy can not only diagnose submucosal cystic adenomyosis but also treat it. For patients with fertility requirements, complete resection of the lesion under hysteroscopy is the first choice. This treatment option can significantly improve symptoms and has a low recurrence rate.

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**Authors' contributions**
Conception and design: Zhenhong Wang and Zhenna Wang. Performed the analyses: Sang Guo. Manuscript writing: Sang Guo. Prepared figure1: Yan Lin. Prepared figure2: Zhenna Wang. Prepared figure3: Zhenhong Wang. Prepared figure4-5: Jianfang Zhu. Prepared table1-5: Sang Guo. Final approval of manuscript: All authors.

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**Availability of data and materials**
The original contributions presented in the study are included in the article, further inquiries can be directed to the corresponding author.

## Declarations

**Ethics approval and consent to participate**
This study, conducted in accordance with the Declaration of Helsinki, received approval from the Ethics Committee of Fujian Maternal and Child Health Hospital (#2024KY039). Consent to Participate declaration: not applicable.

**Consent for publication**
Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

**Competing interests**
The authors declare no competing interests.

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