

## Case Report

## Secondary cystic adenomyosis in a young woman: a case report

Hong-Xuan Li<sup>1</sup>, Ke-Juan Song<sup>2</sup>, Nan-Nan Xia<sup>1</sup>, Xiao-Yu Ji<sup>1</sup>, Lei Sui<sup>1</sup>, Qin Yao<sup>2,\*</sup><sup>1</sup>Department of Medicine, Qingdao University, 266101 Qingdao, Shandong, China<sup>2</sup>Department of Obstetrics and Gynecology, Affiliated Hospital of Qingdao University, 266071 Qingdao, Shandong, China\*Correspondence: [dr\\_yaoqin@126.com](mailto:dr_yaoqin@126.com) (Qin Yao)

Academic Editor: Michael H. Dahan

Submitted: 12 September 2020 Revised: 14 November 2020 Accepted: 18 November 2020 Published: 21 September 2022

## Abstract

**Background:** Cystic adenomyosis (CA) is a rare form of adenomyosis. **Case:** We present for the first time secondary CA in a young woman with increased menstrual volume as the main clinical manifestation. A 23-year-old woman with a history of two uterine surgeries was hospitalized with increased menstrual flow volume and no dysmenorrhea or other discomfort. Ultrasound resulted in misdiagnosis as myoma of the uterus. She underwent laparoscopic surgery for adenomyoma excision with levonorgestrel-releasing intrauterine system (LNG-IUS) placement. During surgery, the lesions were completely removed under ultrasound guidance, and a specimen bag was used to reduce residual lesions in the abdominal cavity. She was postoperative treated with gonadotropin-releasing hormone analog (GnRH-a). Surgical findings and postoperative pathology confirmed CA. She has been followed up for 2 years without recurrence. **Conclusions:** A CA diagnosis should be considered for clinical manifestations of increased menstrual volume and dysmenorrhea in young patients with previous uterine surgery histories. Magnetic resonance imaging (MRI) is very important for CA diagnosis. Conservative surgery along with GnRH-a and LNG-IUS therapy can effectively prevent recurrence.

**Keywords:** Cystic adenomyosis; Adenomyoma excision; GnRH-a; LNG-IUS

## 1. Introduction

Adenomyosis is a common benign gynecologic disease characterized by invasion of the endometrial matrix or glands into the myometrium [1]. The disease may be focal or diffuse [2] and may appear as small cystic spaces, usually <5 mm in diameter, containing a small amount of old blood [3]. When the fused or single lumen of the cystic space has a diameter greater than or equal to 1 cm, the condition is called cystic adenomyosis (CA) [4]. CA is a special type of focal adenomyosis, also called cystic adenomyoma or adenomyotic cyst. Its main clinical manifestation is dysmenorrhea and is most commonly found in younger women (under the age of 30) who have no history of uterine surgery [5].

Consistent with the treatment principle for adenomyosis, the treatment principle for CA is to completely remove the lesion, promote fertility and prevent recurrence [6]. Complete resection of the adenomyoma and restoration of the normal shape of the uterus can prevent recurrence [7]. According to the location and scope of the lesion, different surgical methods can be selected. According to whether the uterus is retained, surgical intervention can involve conservative surgery or total hysterectomy [8]. CA is more common in women of childbearing age, so it is important to preserve fertility. This disease is a special type of adenomyosis, and there may be residual ectopic endometrium after surgery. Therefore, it is particularly important to take appropriate measures to prevent recurrence. We report for the first time a rare case of a CA

secondary to uterine surgery in a 23-year-old woman. Her clinical manifestations were primarily increased menstrual volume without the typical symptom of dysmenorrhea. The patient was successfully treated by the use of conservative surgery combined with gonadotropin-releasing hormone analog (GnRH-a) and a levonorgestrel-releasing intrauterine system (LNG-IUS) for the first time, and no recurrence was found during the 2-year follow-up.

## 2. Case report

A 20-year-old young woman with no history of pregnancy underwent hysteroscopic submucosal myoma resection. Myoma recurrence was found on reexamination 6 months after surgery. Her menstrual volume had slightly increased, and she had painful menstruation. Since then, there had been a small amount of irregular vaginal bleeding and occasional abdominal distension.

At 21 years old, her ultrasound showed a  $4.3 \times 3.4 \times 2.6$  cm heterogeneous echogenic mass on the left wall of the uterine cavity and myometrium, which had been only  $2.0 \times 1.8 \times 1.3$  cm (Fig. 1A) in size according to ultrasonography two months prior. The tumor was close to the serous layer, and multiple nonechogenic masses were distributed within the cavity. The largest nonechogenic mass was  $3.4 \times 1.2$  cm, and a few blood signals were observed inside of the mass (Fig. 1B). The patient was preliminarily diagnosed with uterine myoma degeneration. The patient's hemoglobin was 61 g/L. Considering the rapid growth of the tumor and the anemia caused by the tumor, we per-



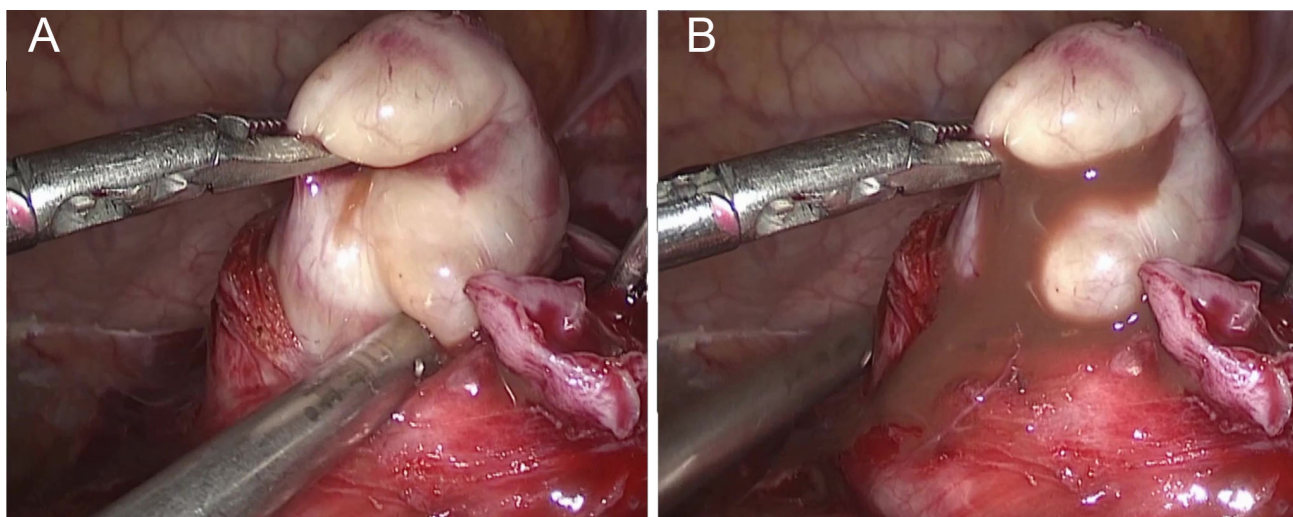


**Fig. 1. Ultrasound tomographs of the patient.** (A) At the age of 21, the patient's first ultrasound examination showed a mass with mixed echogenicity in the uterine cavity of  $2.0 \times 1.8 \times 1.3$  cm in size, with 3–4 echogenicity points and no obvious blood flow signal. (B) At the age of 21, her ultrasonography 2 months later showed a  $4.3 \times 3.4 \times 2.6$  cm heterogeneous echogenic mass on the left wall of the uterine cavity and myometrium (close to the serous layer), with multiple nonechogenic masses distributed within the cavity. The largest nonechogenic mass was  $3.4 \times 1.2$  cm, and a few blood signals were observed inside the mass. (C) The ultrasound demonstrated two hypoechoic nodules in the posterior wall of the uterus,  $4.3 \times 3.3$  cm and  $4.1 \times 3.0$  cm in size; the two nodules were fused and protruded into the uterine cavity.

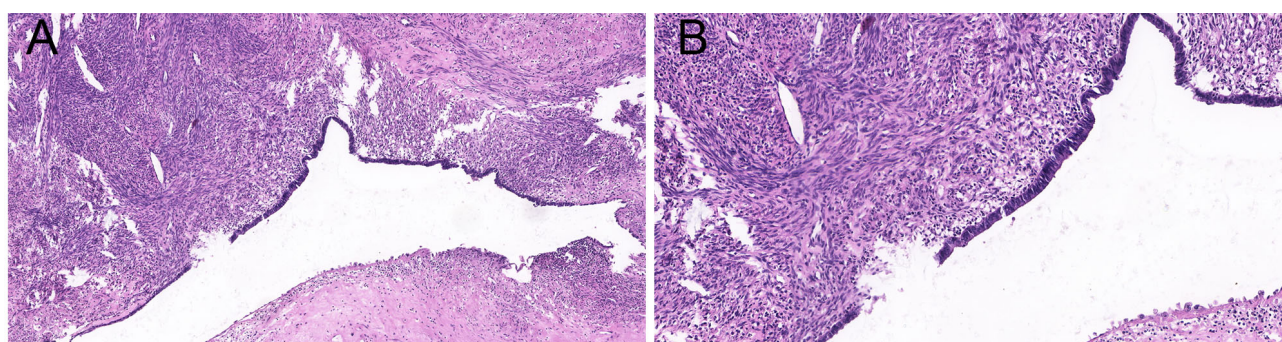
formed a hysteroscopy examination. Intraoperatively, a mass of approximately  $4.5 \times 3.5 \times 3.0$  cm found in the left side wall of the uterine cavity near the angle of the uterus, and its surface was dark red. The base was approximately 3.5 cm wide, the internal protrusion was approximately 70%, the intima was thin, the right fallopian tube opening was visible, and the left fallopian tube opening was unclear. After that, she underwent hysteroscopic surgery. From the free edge of the mass, the mass was cut with a 90-degree circular monopole, and 30 units of oxytocin were used to promote the protrusion of the mass into the uterine cavity. The tumor was found to be soft with an unclear boundary and could be caved in with pressure. The tumor protruding from the uterine cavity was removed. The tumor was approximately 0.5 cm from the serous layer after resection. A small amount of unresectable fibroid tissue with a diameter of approximately 0.5 cm was found under ultrasound guidance. Strict electrocoagulation was used to stem bleeding, and the uterine cavity returned to its normal shape. The postoperative pathology showed adenomyoma of the uterus. One month after the operation, hysteroscopy reexamination showed a normal uterine cavity, with no obvious fibroids in the cavity. The patient was not subsequently re-examined.

At the age of 23, the woman was hospitalized for four months due to increased menstrual flow. Before admission, her menstrual volume had increased over the past 4 months to approximately 1.5 times the original menstrual volume. She had no dysmenorrhea or changes in the menstrual cycle or menstrual period. Ultrasound demonstrated two hypoechoic nodules in the posterior wall of the uterus,  $4.3 \times 3.3$  cm and  $4.1 \times 3.0$  cm in size; the two nodules were fused and protruded into the uterine cavity (Fig. 1C). She was initially diagnosed with submucous uterine myoma. Her hemoglobin was 85.00 g/L. The carbohydrate antigen 125 level was 44.14 U/mL. Considering that the pa-

tient was young, had a desire to have children in the future and that the tumor was the cause of anemia, we treated her with laparoscopic surgery for adenomyoma excision and a levonorgestrel-releasing intrauterine device (LNG-IUD) was inserted. Her uterus had a smooth surface and had increased in size to that of a 2-month pregnancy. On the left side of the uterus, there was a projection of adenomyomatous material with an irregular, lobulated shape and a diameter of approximately 8 cm that contained cystic spaces of 1–2.5 cm in diameter. The cystic cavity contained a chocolate-colored liquid (Fig. 2). Part of the tumor protruded into the uterine cavity, and part extended into the blood vessels of the muscularis. The 1 : 60 diluted pituitrin was injected into the myometrium around the adenomyoma, and the left lateral myometrium was longitudinally incised by unipolar electrocoagulation under the guidance of ultrasound. The adenomyoma was removed by blunt and reticular stripping with a diameter of approximately 8 cm, and the endometrium was penetrated to approximately 1 cm during the operation. The myometrium was continuously sutured with absorbable suture line no. 0. After the tissue was put into a specimen bag and cut with an electromechanical morcellator, the fibroid comminuted tissue was removed from the puncture hole in the right lower abdomen by stages. No residual tissue was detected by ultrasound. The LNG-IUD was placed into the uterine cavity. Postoperatively, she was treated with GnRH-a. The postoperative pathology was adenomyoma (Fig. 3). Immunohistochemistry showed SMA (+), Calretinin (-), CD10 part (+), Caldesmon (+), and ki-67 (+) of approximately 2%. She was followed up for 2 years after the operation, and there was no recurrence.



**Fig. 2.** (A) On the left side of the uterus, there was a projection of adenomyomatous material with an irregular, lobulated shape and a diameter of approximately 8 cm. (B) When the cyst roof was opened, chocolate-colored fluid flowed out of the lesion.



**Fig. 3.** Histologic findings of cystic adenomyosis: hematoxylin and eosin staining: the capsule of cystic adenomyosis is arranged by endometrial glands and the stroma, and around the capsule, the organization has muscular wrapping (A: 50 $\times$  B: 100 $\times$ ).

### 3. Discussion

CA can be divided into juvenile CA (JCA) and adult CA. JCA is a disease that causes severe dysmenorrhea in women less than 30 years of age with no history of uterine surgery, and imaging findings show cystic lesions with a diameter of  $>1$  cm independent of the uterine cavity and covered by the muscular layer [9]. Adult CAs are not well defined and are most common in women over 30 years of age who have a history of previous uterine operations [10].

Injury to the junctional zone (JZ) caused by uterine surgery may be an important factor in the occurrence of adenomyosis [11], resulting in the invasion of the basal endometrium into the myometrium, causing regeneration, healing and changes in the size [12]. We report a rare case of CA in a patient under the age of 30 who had a history of uterine surgery and could not be diagnosed with either JCA or adult CA. She should be considered to have secondary CA associated with surgery. Our patient's secondary adenomyoma may have been associated with JZ injury during myomectomy, leading to endometrial invasion of the myometrium. This hypothesis is supported by the fact that

patients recurred only 6 months after surgery. The second surgery probably resulted in the development of CA. Adenomyoma of the uterus partly protruded from the uterine cavity and partly extended to the muscularis vessels, increasing the possibility of intraoperative JZ injury. Intraoperative ultrasound scans revealed residual tissue from secondary surgery, although intraoperative tight electrocoagulation and postoperative hysteroscopy showed no residuals. The location of the cystic lesion was the same as before, and symptoms of increased menstrual volume appeared. This confirms the possibility of surgical residual tissue or JZ injury.

In the pathology of CA, the cyst wall is composed of endometrial glands and stroma, and the surrounding wall is covered with proliferative fibroid tissue [13]. Most cases of CA are confirmed by postoperative pathology [12]. Our patient was misdiagnosed with uterine fibroids before surgery and diagnosed with CA after surgery. Similar to adenomyosis, CA has nonspecific clinical manifestations, such as dysmenorrhea, chronic pelvic pain, abnormal uterine bleeding, etc. [10]. According to a review, patients under 30



years of age with CA are characterized by drug-refractory dysmenorrhea, whereas those over 30 years of age mainly suffer from chronic pelvic pain or sometimes abnormal uterine bleeding [14]. Dadhwal *et al.* [15] reported two cases of dysmenorrhea in patients under 30 years of age who had no changes in menstrual volume. Pontrelli *et al.* [16] reported a 27-year-old patient with CA who had increased menstrual volume with dysmenorrhea. We reported for the first time a case of CA in a patient under 30 years of age, whose main clinical manifestation was increased menstrual volume without dysmenorrhea.

Ultrasound is the most commonly used imaging method in the diagnosis of adenomyosis, and it can distinguish between adenomyomas and myomas. The accuracy of ultrasound depends on operator experience and expertise and the quality of the equipment [17]. The sensitivity of transvaginal or transanal ultrasound is higher than that of transabdominal ultrasound [18]. Koukoura *et al.* [19] reported a case in which transabdominal ultrasound misdiagnosed a patient with an ovarian cyst, but transanal ultrasound and magnetic resonance imaging (MRI) correctly diagnosed CA. MRI can confirm a diagnosis made by ultrasound [20]. MRI can accurately assess the contents of CA, endometrial and muscular tissue, and the junction area. The fluid in the cysts shows a high signal intensity on T1-weighted images, and the surrounding muscular layer shows low signal intensity on T2-weighted images [10,21]. Hysteroscopy can diagnose adenomyosis according to the special microscopic manifestations [22]. Hysteroscopy can differentiate JCA from congenital developmental abnormalities, such as bicornuate uterus or ovarian cysts, and the lesions can be sampled to obtain pathological specimens for a definitive diagnosis [16]. Alabiso *et al.* proposed a new decision algorithm based on a 28-year-old infertility patient with uterine adenomyosis. First, the diagnosis was made by ultrasound and then confirmed by MRI. Then, hysteroscopy or laparoscopy was performed, and finally, a detailed diagnosis and treatment plan was made [12]. Unfortunately, our patient was misdiagnosed with uterine fibroids by ultrasound, and no further MRI examination was performed, nor was the mass sampled for pathological examination during hysteroscopy. MRI is very important for adenomyosis patients, especially for the identification of recurrent myoma lesions in young patients. Hysteroscopy is also very important for the diagnosis and treatment of intrauterine masses.

Surgery is an important means to treat adenomyosis, drugs can also treat it [23]. Drug treatments include GnRH-a, nonsteroidal anti-inflammatory drugs, LNG-IUS, etc. Drugs can only relieve symptoms, and recurrence can occur after drug withdrawal [24]. There are very few reports about CA drug therapy. Zhou *et al.* [25] reported a case of adult CA patient with significantly reduced symptoms after LNG-IUS treatment. Intraoperative residual lesions or JZ damage may cause recurrence. Surgery may lead to an adverse pregnancy history [26]. According to previous

reports, conservative surgery combined with GnRH-a or an LNG-IUS can prevent recurrence and relieve dysmenorrhea and other symptoms and improve the fertility of patients [27–29]. There is a lack of reliable reports on pregnancy outcomes of CA.

In our patient, her lesion was completely excised by ultrasonic monitoring, and the mass was put into a sample bag and removed from the puncture hole by stages, further reducing residual tissue in the abdominal cavity. GnRH-a and an LNG-IUS were used for postoperative treatment to reduce symptoms and prevent recurrence. We report for the first time the successful treatment of a secondary CA patient under 30 years of age with surgery and medication. This case also has the longest follow-up to date. She was followed up for nearly 2 years without recurrence or obvious discomfort. Her pregnancy outcomes require further follow-up.

## 4. Conclusions

Younger patients with a history of uterine surgery may have secondary CA, and the clinical manifestations may not include the typical unbearable dysmenorrhea. For patients with unclear ultrasound diagnoses, MRI examination is very important.

For patients with intrauterine lesions, hysteroscopy can obtain a sample for pathologic diagnosis or treatment. Complete intraoperative excision of the lesion and reduction in residual tissue, combined with postoperative treatment with GnRH-a and an LNG-IUS, can effectively prevent recurrence and improve prognosis. The prognosis and pregnancy outcomes of CA are still not clear, and further study is needed.

## Abbreviations

CA, Cystic adenomyosis; GnRH-a, gonadotropin-releasing hormone analog; JCA, juvenile adult cystic adenomyosis; JZ, junctional zone; LNG-IUD, levonorgestrel-releasing intrauterine device; LNG-IUS, levonorgestrel-releasing intrauterine system; MRI, magnetic resonance imaging.

## Author contributions

HXL, KJS, NNX and QY participated in the diagnosis and management of this case. XYJ and LS followed up the patient HXL wrote the manuscript. QY revised the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

## Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institution, and written consent was obtained. The study was conducted in accordance with the Declaration of

Helsinki, and the protocol was approved by the Ethics Committee of Affiliated Hospital of Qingdao University (approval number: QYFY WZLL 25965).

## Acknowledgment

Thanks to all the peer reviewers for their opinions and suggestions.

## Funding

This research received no external funding.

## Conflict of interest

The authors declare no conflict of interest.

## References

- [1] Donnez J, Donnez O, Dolmans M. Introduction: Uterine adenomyosis, another enigmatic disease of our time. *Fertility and Sterility*. 2018; 109: 369–370.
- [2] Harada T, Khine YM, Kaponis A, Nikellis T, Decavalas G, Taniguchi F. The impact of adenomyosis on women's fertility. *Obstetrical & Gynecological Survey*. 2016; 71: 557–568.
- [3] Tamai K, Koyama T, Umeoka S, Saga T, Fujii S, Togashi K. Spectrum of MR features in adenomyosis. *Best Practice & Research Clinical Obstetrics & Gynaecology*. 2006; 20: 583–602.
- [4] Cucinella G, Billone V, Pitruzzella I, Lo Monte AI, Palumbo VD, Perino A. Adenomyotic cyst in a 25-year-old woman: case report. *Journal of Minimally Invasive Gynecology*. 2013; 20: 894–898.
- [5] Fan Y, Liu Y, Li J, Fu Y. Intrauterine cystic adenomyosis: report of two cases. *World Journal of Clinical Cases*. 2019; 7: 676–683.
- [6] Kwack JY, Im KS, Kwon Y. Conservative surgery of uterine adenomyosis via laparoscopic versus laparotomic approach in a single institution. *Journal of Obstetrics and Gynaecology Research*. 2018; 44: 1268–1273.
- [7] Kwack J, Kwon Y. Laparoscopic surgery for focal adenomyosis. *Journal of the Society of Laparoendoscopic Surgeons*. 2017; 21: e2017.00014.
- [8] Osada H. Uterine adenomyosis and adenomyoma: the surgical approach. *Fertility and Sterility*. 2018; 109: 406–417.
- [9] Takeuchi H, Kitade M, Kikuchi I, Kumakiri J, Kuroda K, Jinushi M. Diagnosis, laparoscopic management, and histopathologic findings of juvenile cystic adenomyoma: a review of nine cases. *Fertility and Sterility*. 2010; 94: 862–868.
- [10] Gordts S, Grimbizis G, Campo R. Symptoms and classification of uterine adenomyosis, including the place of hysteroscopy in diagnosis. *Fertility and Sterility*. 2018; 109: 380–388.e1.
- [11] García-Solares J, Donnez J, Donnez O, Dolmans M. Pathogenesis of uterine adenomyosis: invagination or metaplasia? *Fertility and Sterility*. 2018; 109: 371–379.
- [12] Alabiso G, Alio L, Arena S, Barbasetti di Prun A, Bergamini V, Berlanda N, *et al.* Adenomyosis: what the patient needs. *Journal of Minimally Invasive Gynecology*. 2016; 23: 476–488.
- [13] Acién P, Bataller A, Fernández F, Acién MI, Rodríguez JM, Mayol MJ. New cases of accessory and cavitated uterine masses (ACUM): a significant cause of severe dysmenorrhea and recurrent pelvic pain in young women. *Human Reproduction*. 2012; 27: 683–694.
- [14] Brosens I, Gordts S, Habiba M, Benagiano G. Uterine cystic adenomyosis: a disease of younger women. *Journal of Pediatric and Adolescent Gynecology*. 2015; 28: 420–426.
- [15] Dadhwal V, Sharma A, Khoiwal K. Juvenile cystic adenomyoma mimicking a uterine anomaly: a report of two cases. *Eurasian Journal of Medicine*. 2017; 49: 59–61.
- [16] Pontrelli G, Bounous VE, Scarperi S, Minelli L, Di Spiezio Sardo A, Florio P. Rare case of giant cystic adenomyoma mimicking a uterine malformation, diagnosed and treated by hysteroscopy. *Journal of Obstetrics and Gynaecology Research*. 2015; 41: 1300–1304.
- [17] Sam M, Raubenheimer M, Manolea F, Aguilar H, Mathew RP, Patel VH, *et al.* Accuracy of findings in the diagnosis of uterine adenomyosis on ultrasound. *Abdominal Radiology*. 2020; 45: 842–850.
- [18] Bazot M, Daraï E. Role of transvaginal sonography and magnetic resonance imaging in the diagnosis of uterine adenomyosis. *Fertility and Sterility*. 2018; 109: 389–397.
- [19] Koukoura O, Kapsalaki E, Daponte A, Pistofidis G. Laparoscopic treatment of a large uterine cystic adenomyosis in a young patient. *BMJ Case Reports*. 2015; 2015: bcr2015210358.
- [20] Kobayashi H, Matsubara S. A classification proposal for adenomyosis based on magnetic resonance imaging. *Gynecologic and Obstetric Investigation*. 2020; 85: 118–126.
- [21] Chua HKA, Goh SYC, Upamali V, Seet MJ, Wong PCA, Phoon WLJ. Subserosal adenomyotic cysts and peritoneal inclusion cysts—Unusual differential diagnoses of multicystic pelvic masses: a review of two cases. *Case Reports in Women's Health*. 2020; 27: e00193.
- [22] Di Spiezio Sardo A, Calagna G, Santangelo F, Zizolfi B, Tanos V, Perino A, *et al.* The role of hysteroscopy in the diagnosis and treatment of adenomyosis. *BioMed Research International*. 2017; 2017: 2518396.
- [23] Lacheta J. Uterine adenomyosis: pathogenesis, diagnostics, symptomatology and treatment. *Ceska Gynekologie*. 2019; 84: 240–246.
- [24] Gao Y, Shan S, Zhao X, Jiang J, Li D, Shi B. Clinical efficacy of adenomyectomy using “H” type incision combined with Mirena in the treatment of adenomyosis. *Medicine*. 2019; 98: e14579.
- [25] Zhou Y, Chen Z, Zhang X. Giant exophytic cystic adenomyosis with a levonorgestrel containing intrauterine device out of the uterine cavity after uterine myomectomy: a case report. *World Journal of Clinical Cases*. 2020; 8: 188–193.
- [26] Sugiyama M, Takahashi H, Baba Y, Taneichi A, Suzuki H, Usui R, *et al.* Perinatal outcome of pregnancy after adenomyectomy: summary of 10 cases with a brief literature review. *Journal of Maternal-Fetal & Neonatal Medicine*. 2020; 33: 4145–4149.
- [27] Oliveira MAP, Crispi CP, Brollo LC, Crispi CP, De Wilde RL. Surgery in adenomyosis. *Archives of Gynecology and Obstetrics*. 2018; 297: 581–589.
- [28] Sun C, Ren X, Gao Y, Liang Z, Mou M, Gu H, *et al.* Clinical efficacy and safety of major uterine wall resection and reconstruction of the uterus combined with LNG-IUS for the treatment of severe adenomyosis. *Geburtshilfe und Frauenheilkunde*. 2020; 80: 300–306.
- [29] Xie M, Yu H, Zhang X, Wang W, Ren Y. Elasticity of adenomyosis is increased after GnRHa therapy and is associated with spontaneous pregnancy in infertile patents. *Journal of Gynecology Obstetrics and Human Reproduction*. 2019; 48: 849–853.