

A case report on cystic adenomyosis: A rare variant of uterine adenomyosis

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Abstract

Background: Cystic adenomyosis is a rare variant of adenomyosis that can easily be misdiagnosed, due to the non-specific nature of symptoms. USS and MRI are essential in making a diagnosis which is later confirmed through histopathological evaluation. Surgical interventions are favored over medical management, as complete resection reduces the risk of recurrences and alleviates the chances of malignant transformation.

Case presentation: A 37-year-old multipara presented with chronic abdominal pain, exacerbated by menstruation, lasting most days of the month for over one year. Clinical evaluation revealed a tender, less mobile pelvic mass which mimicked an intramural fibroid on USS. Considering the long-standing symptoms and lack of fertility wishes, she underwent a laparotomy. During surgery, an unexpected finding of chocolate-colored exudate with a thick epithelium-lined myometrial cystic lesion was noted. Histopathology evaluation revealed fragmented tissue composed of smooth muscle bundles and are lined by endometriotic type epithelium and stroma. There was no malignant changes. Overall findings are in favor of cystic adenomyosis.

Discussion and conclusions: Cystic adenomyosis is considered to arise due to persisting müllerian epithelial cells stimulated by estrogen leading to periodic bleeding into the myometrium. Surgery of the myometrium can also introduce endometrial tissue to the myometrium. To preserve the quality of life and reduce the risk of recurrence, minimal access surgery should be carried out.

Even though rare, a young woman presenting with chronic pelvic pain, and not responding to analgesics should raise the possibility of cystic adenomyosis, which should warrant further investigation and treatment.

Keywords: cystic adenomyosis, myometrial cysts, chronic pelvic pain

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Background

Adenomyosis is a common gynecological pathology where endometrial glands and stroma invade the uterine myometrium. For descriptive purposes, it is further divided into diffuse and focal adenomyosis. Cystic adenomyosis is a rare, sporadic disease characterized by chocolate-like viscous fluid-contained lesions in the myometrium¹. It represents focal adenomyosis. At present, only 47 cases have been reported in the literature, including this case report. Cystic adenomyosis often mimics the natural history of hemorrhagic degeneration of fibroids. Apart from that, congenital malformations and ovarian pathology needs to be kept in mind when making a diagnosis².

This condition is predominately seen in young fertile age group of women and has a varied clinical presentation. Commonly these females seek medical attention for dysmenorrhea, chronic pelvic pain, and abnormal uterine bleeding. There were cases which had been diagnosed incidentally with no symptoms whatsoever.

Case presentation

A 37-year-old multipara female was referred by a general practitioner who was evaluated and treated for chronic lower abdominal pain for almost one year duration. Her obstetric history was rather uncomplicated, where both her children were delivered vaginally with no complications and no history of subfertility. Since the second child, she had been on intramuscular progesterone for nearly two years, after which she had discontinued due to weight gain. Her menstrual cycles were regular with 3-5 days of bleeding, and except

for dysmenorrhea outlasting cycles, no other menstrual symptoms had been noted. In this background, she had developed a non-specific lower abdominal pain with an insidious onset, lasting for most days of month, which had progressively worsened over time and had not responded to analgesics.

On abdominal and pelvic examination, a tender uterine origin pelvic mass with reduced mobility of the size of a 16 weeks' gravid uterus was felt. Ultrasonography revealed a 8.2×7.3 cm pelvic mass, which was suggestive of an intramural fibroid on the anterior uterine wall. Her preliminary blood investigations were normal and urine HCG was negative.

Considering her symptoms and ultrasonic appearance of the mass, patient was counselled for surgical intervention. However, she insisted on uterine preservation but requested a simultaneous tubal sterilization.

During surgery, an unexpected finding of chocolate-colored exudate with thick epithelium-lined myometrial cystic lesion was noted. Following drainage of altered blood, excision of epithelial lining was done and the uterine wall repaired. Intentionally the endometrial cavity was opened into, to exclude any concomitant or extension of the pathology. For histopathological purposes, epithelium lined myometrial segments were sent as per laboratory guidelines.

Histopathology evaluation revealed fragmented tissue composed of smooth muscle bundles and are lined by endometriotic type epithelium and stroma. There was no malignant changes. Overall findings are in favor of cystic adenomyosis.

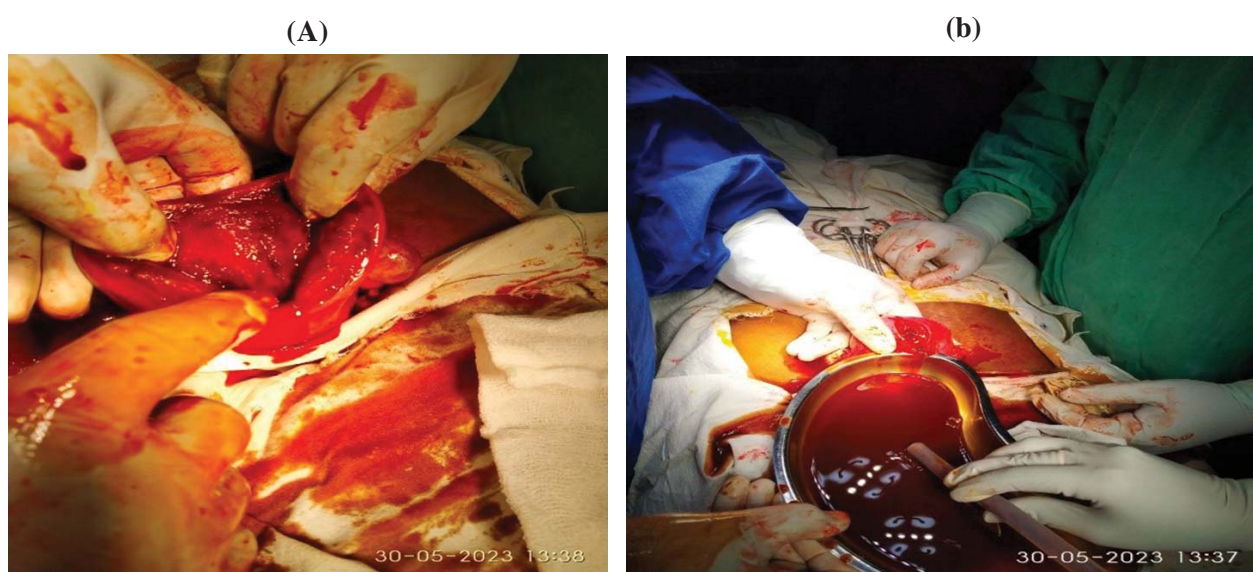


Figure 1. (Image-A&B: shows chocolate colored fluid release with myometrial incision and cystic space within myometrium).

Discussion

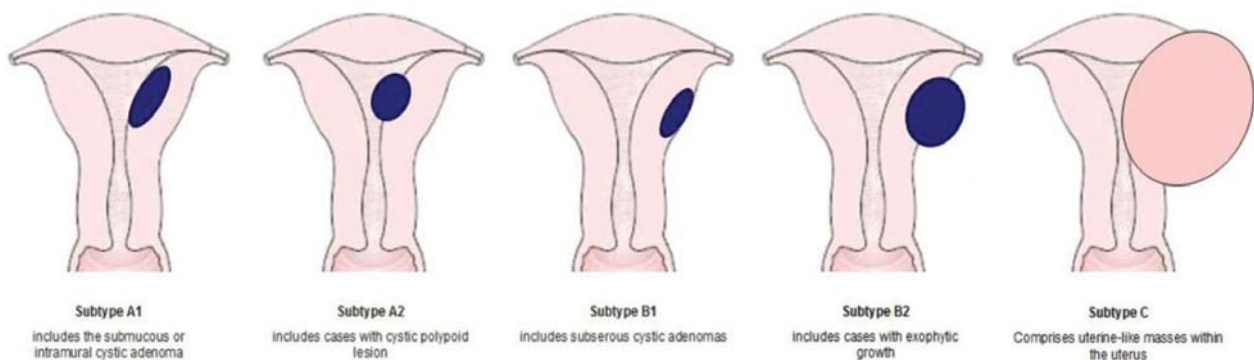
According to age of presentation, cystic adenomyosis is further classified as primary (juvenile) or secondary cystic adenomyosis. Primary disease manifest about five years after menarche (around 18-years) whereas secondary disease appears after 30-years of age.

Depending on location, cystic adenomyosis is further classified² as –

- **A1 subtype- submucosal or intramural cystic adenomyosis**
- **A2 subtype-cystic polypoid lesions**
- **B1 subtype-subserosal cystic adenomyosis**
- **B2 subtype-cases of intrauterine growth**
- **C subtype-similar cysts in the uterus**

Most cystic adenomyosis appears to be intra myometrial and very few are sub-serosal^{1,10}.

Classification of Adenomyotic Cysts Proposed by I. Brosens



<https://hysteroscopynewsletter.com/2019/08/07/hysteroscopic-treatment-of-submucous-cystic-adenomyosis/>

The pathogenesis of cystic adenomyosis is unclear. There are two proposed mechanisms are noted in literature¹. First hypothesized mechanism is thought to be due to persisting mullerian epithelial cells which reacts to estrogen causing periodic bleeding inside the myometrium. The other method is considered to be iatrogenic, uterine surgeries before onset of disease leading to dispersion of endometrial tissues into myometrium.

Diagnosis of cystic adenomyosis is supported by ultrasonography and MRI scans. MRI being the most sensitive³. MRI will show high signal intensity on the T1-weighted image and significantly low signal intensity on the T2-weighted image³.

Sophisticated investigations apart, what is most important is the clinical suspicion that a young woman

presenting with severe dysmenorrhea or unresolving chronic pelvic pain could be due to this rare yet important clinical entity of cystic adenomyosis. In certain cases, to exclude other differential diagnoses, such as unicornuate uterus with fluid filled rudimentary horn there is a need of diagnostic hysteroscopy of hysterosalpingography⁷.

Diagnostic criteria for cystic adenomyosis includes⁴ (adapted from Zhao CZ et al)

- Isolated lesion
- No abnormalities in the uterus, fallopian tubes, and ovaries
- Post-operative lesions with pathological reports of cystic adenomyosis
- Excised lesions with endometrial glands and interstitium

- Lesions that contain viscous chocolate-like liquid
- Small lesions of adenomyosis, such as adenomyoma, adjacent to cystic lesion

Apart from pain and bleeding symptoms affecting quality of life, there is a risk of secondary malignant transformation causing risk to life⁵. This signifies the need of active intervention once condition is diagnosed. Age, symptoms, size of cyst, malignancy status and fertility wishes influence management options. The main aims of treatment are to remove the cyst, to improve fertility, prevent recurrences and to minimize the risk of malignancy.

Conservative / medical management is suitable for women with mild symptoms and smaller cysts. Therapeutic options include, standard analgesics therapy, hormonal suppression using COCP, LNG-IUS or GnRH agonists. The downside for medical management include the possibility of relapsing once treatment is halted and the interference with fertility wishes.

Surgery remains the mainstay of management. Open, laparoscopic or hysteroscopic approaches can be used and whenever feasible, minimal access surgical interventions must be considered as it has many advantages over open surgery⁹. Hysteroscopy avoids abdominal incisions, serosal and outer myometrial incisions. But larger lesions are difficult to manage by this route⁶. When no fertility wishes and if patient consented for hysterectomy, surgery itself is curative. Otherwise, complete resection of cyst with wall and adjacent myometrium is the option. There are many other described interventions and attempted interventions including high frequency ultrasound ablations⁸ and radio-frequency ablations with alternating efficacy.

We chose the traditional open surgical approach as complete excision is more promising and post operatively patient was placed on continuous oral progestogen with 6 weeks review plan.

Conclusions

Cystic adenomyosis is a distinct uterine pathology that needs to be kept in mind when evaluating a myometrial cyst in a younger woman. Though rare, it has a potential for malignant transformation. Surgical management is preferred over medical management in

most instances. However lesion size, location, patient age and fertility wishes should factor-in when deciding the treatment modality.

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Conflicts of interests

None.

References

1. Xu T, Li Y, Jiang L, Liu Q, Liu K. Subserous Cystic Adenomyosis: A Case Report and Review of the Literature. *Sec. Obstetric and Gynecological Surgery* 2022; 9. <https://doi.org/10.3389/fsurg.2022.807676>
2. Brosens I, Gordts S, Habiba M, Benagiano G. Uterine Cystic Adenomyosis: A Disease of Younger Women. *Journal of Pediatric and Adolescent Gynecology* 2015; 28(6): 420-6. doi: 10.1016/j.jpog.2014.05.008
3. Tamai K, Koyama T, Umeoka S, Saga T, Fujii S, Togashi K. Spectrum of MR features in adenomyosis. *Best Pract Res Clin Obstet Gynaecol.* 2006; 20: 583. doi: 10.1016/j.bpogyn.2006.01.009
4. Zhao CZ, Wang B, Zhong C, Lu ST, Lei L. Management of uterine cystic adenomyosis by laparoscopic surgery: case report. *BMC Womens Health* 2021; 21: 263. doi: 10.1186/s12905-021-01341-1
5. Gomez NF, Still MA, Akki AS, Carbajal-Mamani SL, Cardenas-Goicoechea J. Uterine clear cell carcinoma arising from cystic adenomyosis: a case report. *J Obstet Gynaecol.* 2021; 41: 1175-7. doi: 10.1080/01443615.2020.1847056
6. Giana M, Montella F, Surico D, Vigone A, Bozzola C, Ruspa G. Large intramyometrial cystic adenomyosis: a hysteroscopic approach with bipolar resectoscope: case report. *Eur J Gynaecol Oncol.* 2005; 26: 462-3.
7. Iain N, Goel S. Cystic Adenomyoma simulates

- uterine malformation: a diagnostic dilemma: case report of two unusual cases. *J Hum Reprod Sci.* 2012; 5: 285-8. doi: 10.4103/0974-1208.106342
8. Zhou XJ, Zhao ZM, Liu P, Zhao CY, Lin YJ, Liu Y, et al. Efficacy of high intensity focused ultrasound treatment for cystic adenomyosis: a report of four cases. *Ann Palliat Med.* 2020; 9: 3742-9. doi: 10.21037/apm-20-1599
 9. Takeda A, Sakai K, Mitsui T, Nakamura H. Laparoscopic management of juvenile cystic adenomyoma of the uterus: Report of two cases and review of the literature. *J Minim Invasive Gynecol.* 2007; 14: 370.
 10. Neri A, Bahary C, Eckerling B, Lurie M. Serosal (subperitoneal) cysts of the uterus. *Am J Obstet Gynecol.* 1968; 102: 612.