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CASE REPORT

A Rare Case of Multiple Subserous Uterine Adenomyomas Misdiagnosed as an Ovarian Cyst, Diagnosed and Treated by Laparoscopy

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Objective: To report a rare case of multiple subserous uterine adenomyomas diagnosed and treated by laparoscopy.

Case Report: A premenopausal 55-year-old woman was admitted presenting with bilateral adnexal cysts. Preoperative ultrasound and magnetic resonance imaging both indicated a right ovarian cyst. However, during the laparoscopic surgery, it was identified that the cyst was multiple subserosal uterine cystadenomas, which were located on the posterior wall of the uterus and adjacent to the right ovary. Postoperatively, the pathology revealed that it was a subserosal uterine cystadenoma.

Conclusion: Subserous adenomyomas, a rare subtype of uterine adenomyomas, are commonly reported in the literature as a single adenomyoma. Furthermore, no studies have reported the presence of multiple subserosal adenomyomas. This condition requires attention, and it is essential to differentiate it from ovarian cysts. Subserosal adenomyomas exceeding 8 cm in diameter are rare, with the literature documenting a mere six cases. Larger cysts are associated with a higher likelihood of malignancy. There is currently no effective drug treatment available for this disease. Laparoscopy is an effective method for treating this condition.

Keywords: adenomyoma, adenomyosis, giant cystic adenomyoma, hysteroscopy

Introduction

Adenomyosis is a benign condition of the uterus. It is characterized by the presence of endometrial glands and stroma within the myometrium, accompanied by adjacent smooth muscle hyperplasia. When a cyst-like space is occupied by misplaced endometrial tissue and blood, measuring over 10 mm in diameter, it is known as cystic adenomyosis. This condition, also known as an adenomyotic cyst or cystic adenomyoma, is a rare type of adenomyosis.[1] Most case reports indicate that the lesion site for uterine cystic adenomyosis is commonly found within the myometrial wall or the subserosal layer.[2] It is a cystic lesion that does not communicate with the uterine cavity and is lined with healthy endometrium, a condition first proposed by Cullen in 1904.[3] Cystic adenomyosis, due to its rarity, often presents challenges in differential diagnosis.[4] Since it is typically located subserosally in the uterus, it is necessary to differentiate it from subserosal uterine fibroids. Unlike subserosal fibroids, it often contains hemorrhagic fluid. There are also documented cases in the literature where subserosal uterine adenomyomas have been misdiagnosed as uterine malformations.[5] When a subserosal adenomyoma is located near the ovary, it is necessary to differentiate it from ovarian tumors.

Some individuals with cystic adenomyosis experience intense menstrual pain, while others may be diagnosed during routine gynecological examinations without presenting any symptoms. The occurrence of dysmenorrhea in women with cystic adenomyomas of the uterus can be attributed to the gradual enlargement of the cyst due to recurrent intra-cystic bleeding during menstruation. As intra-cystic bleeding increases, dysmenorrhea symptoms worsen, rendering analgesics and oral contraceptive pills ineffective. Hormonal therapy with GnRH analogs has been documented, typically showing

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limited efficacy.[6,7] Surgical treatment of patients with symptomatic cystic adenomyoma by the laparoscopic or hysteroscopic route is currently considered an effective method.[8] This article presents a clinical case of a 55-year-old woman with cystic adenomyoma. She was admitted for the evaluation of bilateral adnexal cysts identified during a routine physical examination. Ultrasound and pelvic MRI both indicated a right ovarian cyst. Laparoscopy confirmed the diagnosis of subserosal cystadenoma. The diagnosis and treatment of her condition are discussed to provide a reference for clinicians in diagnosing and treating this disease.

Presentation

We report the case of a 55-year-old woman in the premenopausal stage, who was admitted for the evaluation of bilateral adnexal cysts identified during a routine physical examination. The patient's medical history revealed three natural births and three miscarriages. Ultrasonography unveiled a 7.9 cm × 5.9 cm × 4.8 cm ovarian cyst in the left ovary (Figure 1A and B), accompanied by a 3.4 cm × 2.7 cm × 2.1 cm hydrosalpinx in the right ovary. In the ultrasound image, the cyst presents as a hypoechoic, unilocular structure with a thin wall and suboptimal internal acoustic transmission. Minimal peripheral blood flow signals are detectable Figure 1B shows the ovarian echoes adjacent to the cyst. According to the IOTA standard, the cyst is classified as a B1. Pelvic MRI reveals a cystic lesion measuring approximately 7.5 cm x 4.4 cm x 4.9 cm in the left posterior aspect of the uterus, exhibiting a cast-like distribution. Post-enhancement, the cyst wall shows increased signal intensity (Figure 2). The morphology of the left ovary appears normal with no significant abnormal signal detected. Pelvic ultrasound was often performed before MRI. Given the gynecological ultrasound's suggestion of a pelvic mass potentially originating from the left ovary and the pelvic MRI indicating no abnormalities in the left ovary, with the pelvic mass located posteriorly to the left side of the uterus, we hypothesized that the pelvic mass was situated in the left adnexal region and considered it to be a left ovarian cyst. The patient's serum levels of cancer antigen (CA)-125 and HE4 fell within the normal range.

Due to the inconclusive diagnosis and the large size of the cyst, we recommended that the patient undergo laparoscopic exploration, with intraoperative rapid pathology to preliminarily determine the nature of the lesion. During the subsequent laparoscopic surgery, meticulous exploration revealed the presence of two cystic structures situated along the posterior uterine wall. The larger cyst measures approximately 8 centimeters in diameter (Figure 3A and B), while the smaller one has an approximate diameter of 3 centimeters (Figure 3C). These two cysts are closely juxtaposed and are in immediate proximity to the right ovary (Figure 3D). Notably, there were no discernible adhesions to the surrounding tissues, and chocolate-like viscous liquid was detected inside the cyst. No abnormalities were observed in the bilateral ovaries or fallopian tubes. Additionally, laparoscopy revealed no foci in the pelvic peritoneum).

During the intraoperative period, a consultative discussion was conducted with the patient's family regarding the advisability of performing a total hysterectomy. Despite the medical recommendation, the patient's family expressed their objections. Consequently, the surgical intervention was confined to the excision of the two cystic adenomyomas located

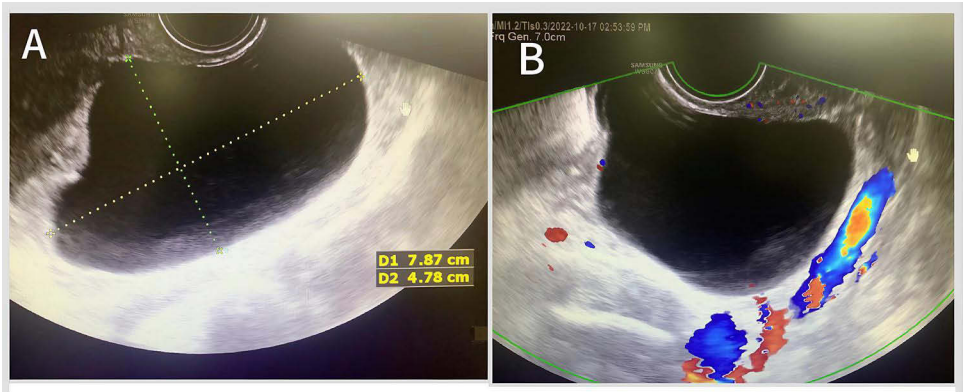

Figure 1 Ultrasonic imaging of the large cystic adenomyoma (**A** and **B**). (**A**) The ultrasound images revealed a cystic mass measuring 7.87 cm by 4.78 cm, which appeared as a hypoechoic area and situated in the retro-left uterine region. (**B**) The cyst presents as a hypoechoic, unilocular structure with a thin wall and suboptimal internal acoustic transmission. Minimal peripheral blood flow signals are detectable. The ovarian echoes adjacent to the cyst.

Figure 2 MRI imaging of the large cystic adenomyoma.

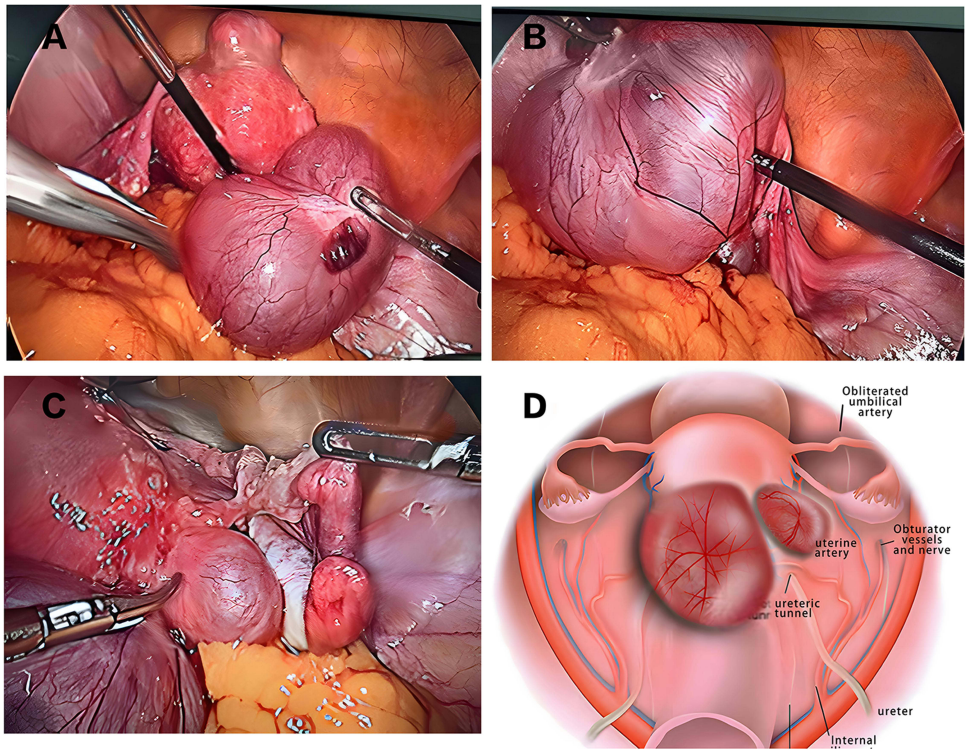

Figure 3 Under laparoscopic vision. (**A** and **B**) The larger cyst measures approximately 8 centimeters in diameter, is located in the posterior lower segment of the uterus. (**C**) The smaller one has an approximate diameter of 3 centimeters. (**D**) These two cysts are closely juxtaposed and are in immediate proximity to the right ovary.

on the posterior aspect of the uterus. The surgical procedure was executed successfully, and the patient exhibited an optimal postoperative recovery. The patient was discharged on the third postoperative day. Pathohistological examination confirmed the diagnosis of uterine cystic adenomyoma, characterized by active epithelial proliferation in the lesion area, surrounded by proliferating smooth muscle tissue (Figure 4).

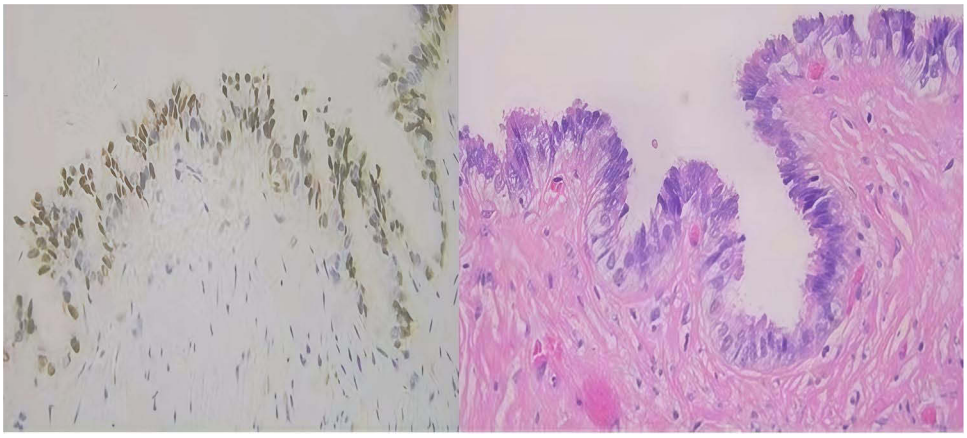

Figure 4 Pathohistological examination confirmed the diagnosis of uterine cystic adenomyoma, characterized by active epithelial proliferation in the lesion area, surrounded by proliferating smooth muscle tissue.

Discussion

Uterine cysts are classified into 2 main groups: congenital and acquired. Congenital cysts of the uterus are divided into Müllerian duct cysts (usually located on the midline) and Wolffian duct cysts (generally situated in the lateral uterine wall). These congenital cysts are filled with clear fluid and are typically unilocular. Acquired cysts include cystic degeneration of uterine leiomyoma, cystic adenomyosis, and serosal cysts.[9,10] The etiology of cystic adenomyoma remains uncertain and may stem from cycles of endometrial injury and repair, elevated estrogen levels, immune responses, inflammatory processes, and other contributing factors.[11] Acién et al criteria for the diagnosis of cystic adenomyosis include (1) isolated accessory mass, (2) normal uterus (endometrial lumen), with normal Fallopian tubes and ovaries, (3) pathological examination of the surgically excised mass, (4) an accessory cavity lined by endometrial epithelium with glands and stroma, (5) a chocolate-brown-coloured fluid content, and (6) no adenomyosis (if the uterus has been removed), although there could be small foci of adenomyosis in the myometrium adjacent to the accessory cavity.[12] In our case, the patient fulfilled all the aforementioned criteria. The histopathological results indicated that the cyst wall was lined with either single-layered or stratified glandular epithelium, with active epithelial proliferation observed in focal areas and hyperplasia of the surrounding smooth muscle tissue. Morphologically, these features are most consistent with a subserosal uterine adenomyoma. Since subserous cystic adenomyosis may not exhibit classic symptoms such as dysmenorrhea, menorrhagia, or chronic pelvic pain, it is prone to misdiagnosis. Pelvic ultrasound is frequently employed as a primary diagnostic tool for women experiencing menstrual irregularities and pelvic discomfort. In this case, a routine health check-up ultrasound examination revealed a cyst in the patient's left adnexal region. Due to the cyst being separate from the uterus and having a thin cyst wall, ovarian echoes were detected around it, which led the sonographer to diagnose it as an ovarian cyst. Giovanni Pontrelli et al have documented a case of cystic adenomyoma, which closely resembles a uterine malformation.[5] Additionally, some cases may be erroneously diagnosed as subserous leiomyomas or broad ligament fibroid.[12,13] MRI appears to be beneficial for patients with suspected adenomyosis when ultrasound findings are inconclusive. Cysts exceeding 8 cm in size are very rare, with only 6 cases reported previously.[14–18] Among these, a duo of cases exhibited neoplastic progression, one transmuting into clear cell carcinoma[16] and another into endometrioid adenocarcinoma.[17] Based on this, it can be deduced that the greater the size of such cysts, the higher the probability of malignant transformation.

The rarity of this diagnosis accounts for the absence of a current consensus on the optimal treatment for patients with cystic adenomyoma. Hormonal therapy, including GnRH agonists or oral contraceptives, has been utilized as a treatment option for cystic adenomyosis and has shown some effectiveness. However, after stopping the medication, symptoms may reappear.[10] Oral contraceptives were ineffective when the cysts were too large.[5] A case report documented an attempt to treat this condition with iterative punctures, which ultimately ended in failure.[19] Surgical treatment of patients with symptomatic cystic adenomyoma by the laparoscopic or hysteroscopic route is currently considered an effective

method. The choice of surgical method is determined by the location and size of the lesion.[8] Operative management poses no unusual challenges, given that the procedures employed are analogous to those used in myomectomy. Since no other endometriotic lesions were found in the pelvis and the patient's family refused to consent to a hysterectomy, only the subserosal cystadenoma of the uterus was excised.

Conclusion

Subserosal uterine adenomyoma is rare, and multiple subserosal adenomyomas are even rarer, with no previous reports in the literature. Ultrasound is an effective diagnostic method, but there is a potential for misdiagnosis. When ultrasound diagnosis is uncertain, MRI can be utilized for further examination. Currently, there are no specific medications available to treat this disease. Combined oral contraceptives and GnRH-a drugs show some effectiveness, but symptoms tend to recur after discontinuation. Surgery is an effective treatment for this condition, with the specific surgical method determined by the location and size of the tumor.

Ethical Approval

The patient signed an informed consent statement. This study was approved by the ethics committee of Hangzhou Women's Hospital. We have obtained institutional approval from the hospital to publish this case in the journal ([2021] A- (7) −05).

Declaration of Patient Consent

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

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Disclosure

All authors have no conflicts of interests to declare.

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