Robot-assisted laparoscopic adenomyomectomy of adenomyotic nodule implanted in the uterine endometrium manifesting as endometrial cancer: a case report and literature review

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Summary
Thickened uterine endometrium with abnormal uterine bleeding highly suggests endometrial hyperplasia or endometrial carcinoma. A case of 35-year-old nulliparous woman came to our department with endometrial mass manifesting as endometrial cancer. Transrectal ultrasonography and magnetic resonance imaging (MRI) showed an 8×6 cm multicystic, ill-defined mass compacted at the uterine endometrium, the anterior wall of the uterus, and 3×3 cm heterogeneous mass at the left adnexa. The endometrial mass showed multiple septations with enhancement and low-signal intensity on T2-weighted images. After endometrial biopsy was done and simple hyperplasia without atypia was observed at the histopathologic finding, the patient underwent robot-assisted laparoscopy and diagnosed as adenomyoma at the frozen pathology. After adenomyomectomy, permanent pathologic analysis revealed the same result and she recovered without any complications and responded well to gonadotropin-releasing hormone (GnRH) agonist therapy.

Key words: Adenomyoma; Robot-assisted laparoscopic surgery; Adenomyomectomy.

Introduction
Uterine adenomyosis is a common benign gynecologic disease, characterized by the presence and growth of heterotopic endometrial or endometrium-like structures in the myometrium [1]. The disease appears in two forms, the diffuse type and the focal type, and the latter is known as adenomyoma [2]. The definitive treatment for adenomyosis is hysterectomy. However, to conserve fertility, treatment options include medical treatments for improving symptoms or hormone therapy with a gonadotropin-releasing hormone (GnRH) agonist, or oral contraceptives. Another option is the excision of the adenomyotic lesion by laparotomy or laparoscopy. There are several conservative surgical methods; however, because of the risks of recurrence of adenomyosis and uterine rupture during pregnancy, safer methods are needed to completely resect adenomyotic tissues.

Herein, the authors present an unusual case of adenomyoma implanted in the uterine endometrium manifesting as endometrial cancer and successful robot-assisted laparoscopic adenomyomectomy. This report may provide informative data for the diagnosis and treatment of this kind of extraordinary disease.

Case Report
A 34-year-old woman, nulligravida, was referred to the present outpatient clinic with symptoms suggestive of endometrial cancer. She visited a local clinic for irregular menstruation and menorrhagia, and transrectal ultrasonography demonstrated thickened endometrium. Therefore, she underwent a magnetic resonance imaging (MRI) test, and endometrial cancer was highly suggestive. Thus, she was referred to the present clinic for further evaluation.

On initial examination, the patient’s height and weight were 173 cm and 88 kg, respectively (body mass index = 29.4 kg/m²). She had experienced menarche at the age of 14 years, and her menstrual cycle was irregular (30–60 day intervals). She had experienced menorrhagia, and her last menstrual period was a month ago, during which vaginal bleeding persisted for a month.

Physical examination demonstrated normal mature female appearance. Speculum examination could not be performed because she was virgin. Transrectal ultrasonography showed an 8×6 cm hyperechoic lesion in the endometrium, and a 4×2.5 cm mass in the left adnexal region. Outside MRI imaging was re-interpreted thoroughly, and it demonstrated an 8×6 cm multicystic, ill-defined mass compacted at the uterine endometrium, in the anterior wall of the uterus, and a 3×3 cm heterogeneous mass at the left adnexa. The endometrial mass showed multiple septations with enhancement and low-signal intensity on T2-weighted images. The official reading was endometrial hyperplasia with suspicious endometrial cancer in the anterior half of the mass. There was no sign of diffuse uterine adenomyosis and no other metastatic lesions were found (Figure 1).
Figure 1. — A: Transrectal ultrasonographic findings of the uterine mass. There is an 8×6 cm multicystic, ill-defined mass compacted in the uterine endometrium. B: Magnetic resonance imaging shows multiple septations with enhancement and low-signal intensity of endometrial mass on T2-weighted images.

Figure 2. — Operative findings. A-C: Vertical incision of the uterus and large adenomyotic nodule is observed. D: After extracting the mass. E-G: Uterine reconstruction with multiple-layered suture. H: After completion of reapproximation of the uterine defect.

The results of the routine laboratory tests, including complete blood count and blood chemistry test, were normal, and the tumor marker levels revealed the following: a carcinoembryonic antigen (CEA) level of 1.98 ng/ml, a cancer antigen 125 (CA-125) level of 14.4 U/ml, and a β-human chorionic gonadotropin (β-hCG) level of 12.85 mIU/ml. She underwent dilatation, curettage, and biopsy to rule out endometrial cancer. The histopathologic result of the endometrial biopsy was simple hyperplasia without atypia. Hysterectomy was recommended for the patient. However, she wished to preserve her fertility, and despite the risk of spreading cancer, she decided to undergo robotic mass excision and confirm the permanent pathologic analysis and undergo reoperation later, if needed.

After providing informed consent, robot-assisted laparoscopy was performed. With the patient in the lithotomy position under general anesthesia, pneumoperitoneum was established by CO₂.
gas insufflations, and four ports, including an accessory one, were inserted with a robotic system camera placed to confirm atraumatic placement of the port.

The uterine corpus was large, and the anterior aspect of the uterus was globular due to the mass (Figure 2). A four-cm mass was observed in the left ovary, and the bilateral fallopian tubes and right ovary appeared to be normal. There were no other abdominal abnormalities or ascites. Depending on the orientation of the mass in preoperative imaging studies, a vertical incision was made on the anterior aspect of the uterus. Along with layer-by-layer dissection of the myometrium, there were several endometrial implantations in the myometrium, which formed an adenomyotic nodule. There was no diffuse adenomyosis and the isolated mass was observed in the anterior wall of the uterus, implanted in the endometrium. The tumor was excised and sent for frozen pathology and the result was adenomyoma (Figure 3). After complete resection of the tumor, the vertical uterine incision was repaired in multiple layers with the use of barbed sutures (1-0 and 2-0 V-LOC). The patient recovered well without any complications and the permanent pathologic analysis revealed the same result with left ovarian dermoid cyst. Outpatient transrectal ultrasonography was performed, and a normal uterus was observed (Figure 4). The patient is now receiving progesterone therapy and GnRH agonist therapy for endometrial hyperplasia and adenomyosis, respectively, and long-term follow-up is planned.

Discussion

Adenomyosis is a common cause of dysmenorrhea and the average age of symptomatic women is usually older than 40 years. In the past, diagnosis relied on clinical manifestations, most frequently abnormal uterine bleeding (50%), secondary dysmenorrhea (30%), and an enlarged, tender uterus [3]. The most commonly affected site is the posterior wall, but any site can be involved [4].

Although clinical diagnosis is important in adenomyosis, imaging studies, including pelvic ultrasound or MRI, can be helpful. The characteristic finding of transvaginal ultrasonography is the presence of three or more specific criteria: heterogeneity, increased or decreased echogenicity, and anechoic lacunae or myometrial cysts. Myometrial cysts in a poorly defined area with an abnormal echo texture are highly specific for adenomyosis, but are only present in 40–60% of cases. When cysts are not present, hypoechoic and heterogeneous regions may be helpful in making a diagnosis [5–7]. As in MRI, typical adenomyosis appears as an ill-demarcated low-signal-intensity area on T2-weighted images due to abundant smooth muscle proliferation. Adenomyoma may appear as an intracavitary, subserosal polypoid mass, or intramyometrial mass and may show heterogeneous signal intensity on T2-weighted images, like adenomyosis. Another atypical morphologic feature of adenomyosis is the pseudo-widening of the endometrium. On T2-weighted images, the endometrial junction may be obscured due to high-signal-intensity linear striations radiating from the endometrium to the area of adenomyosis [8].

The treatment of adenomyosis should consider the patient’s age and disease severity. Medical treatments include symptomatic treatment, such as the administration of nonsteroidal anti-inflammatory drugs and hormone therapy with a GnRH agonist and oral contraceptives. However, its effect is often transient and it is frequently used as preoperative adjuvant therapy or postoperative therapy. There-
fore hysterectomy remains a most important therapeutic option for women with symptomatic adenomyosis [9]. However, women who wish to preserve their fertility, but cannot manage their symptoms with medical therapy, should consider conservative treatment. Adenomyomectomy refers to the surgical excision of myometrial tissue affected by adenomyosis. It was first performed as metroplasty following the repair of a wedge-shaped defect in the myometrium by Hyams in 1952 [10]. Since then, various surgical methods have been introduced. Initial reports of surgical techniques are mostly performed by laparotomy or mini-laparotomy. Unlike uterine myoma, adenomyoma is usually ill-demarcated and often invades the entire uterine muscle layer, hence it was considered that open surgery is more beneficial to radically remove the affected tissue. The first case reports treated by laparoscopy were published in 2004, including three patients [11], and the first case report of robot-assisted excision of a cystic adenomyoma in an adolescent girl was published in 2010 [12]. The benefits of laparoscopic surgery are widely known, with fewer complications, less postoperative pain, a shorter length of surgery, and a shorter hospital stay compared with open surgery. However, it was not preferred by many surgeons because it was thought that in laparoscopic adenomyomectomy, it is difficult to radically resect the affected tissues and it requires highly skilled surgical techniques, including extensive suturing, and the operation time could be even longer than open surgery due to the difficulty of adenomyoma removal and uterine reconstruction. Following that, it may cause incomplete resection of the tumor and extensive intraoperative bleeding. However, imaging techniques are developing progressively and it is possible to predict the precise extent of the adenomyoma with MRI or 3D transvaginal ultrasound. Additionally, one of the advantages of robot-assisted surgery is meticulous suturing. In addition, the benefits of robot-assisted laparoscopic operation are obvious in terms of the cosmetic outcome.

There are non-surgical options, such as uterine artery embolization (UAE) and magnetic resonance-guided focused ultrasound. A review of the management of subfertile women with uterine adenomyosis and adenomyoma reported that there is more evidence supporting the advantages of conservative surgery or UAE for effective symptom control and a longer duration of effectiveness compared with medical treatment alone. Furthermore, a longer cumulative pregnancy rate and higher cumulative final successful delivery rate were observed in women who underwent conservative surgery compared with GnRH agonist treatment [13].

Unlike open adenomyomectomy or even laparoscopic myomectomy, there are just a few cases of conservative laparoscopic adenomyomectomy, especially by robot-assisted laparoscopic adenomyomectomy, and there is little data on indication and surgical technique. As several cases of postoperative uterine rupture were reported after conservative adenomyomectomy, skillful surgical technique is needed to totally remove the uterine tumor to prevent disease recurrence, together with meticulous uterine reconstruction to prevent uterine rupture in future pregnancies. Further investigation on operative procedures and postoperative management of robot-assisted laparoscopic adenomyomectomy is necessary.

In summary, the authors report a rare case of an adenomyotic nodule implanted in the uterine endometrium manifesting as endometrial cancer and introduce robot-assisted laparoscopic adenomyomectomy. Diagnosis is difficult when imaging studies reveal atypical findings of adenomyoma and surgeons should consider conservative surgical treatment, especially when the patient wishes to preserve her fertility.

References


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