Laparoscopic Diagnosis of Adenomyosis : a Case Report

Akira FUJISHITA, Hideaki MASUZAKI, Raúl ORTEGA CHAVEZ, Hisayoshi Nakajima, Tadayuki Ishimaru

Department of Obstetrics and Gynecology, Nagasaki University School of Medicine

Usually, Adenomyosis is diagnosed after surgery. Laparoscopic diagnosis may have advantages over other surgical procedures, especially for infertility patients undergoing expectant therapy. We successfully diagnosed adenomyosis using a laparoscopic needle biopsy technique in an infertility patient.

Key words : adenomyosis, laparoscopy, gonadotropinreleasing hormone agonist (GnRH-a), MRI.

Introduction

Adenomyosis is the invasion and growth of normal endometrium in the myometrium. It is characterized by enlargement of the uterus, dysmenorrhea and menorrhagia and often occurs in the patient's fifties. Traditionally, total hysterectomy has been the treatment of choice. However, women who want to remain fertile often request an alternative treatment for adenomyosis, such as Gonadotropin-releasing hormone agonist (GnRH-a) therapy.

Most cases of adenomyosis are diagnosed from a removed uterus. There are only four cases of conservative treatment in which a definitive diagnosis of adenomyosis was established histologically¹⁴⁾. Laparotomy was performed in all previous cases, but the present case was successfully diagnosed using laparoscopic needle biopsy. To the best of our knowledge, this is the first case report of adenomyosis confirmed by laparoscopical needle biopsy.

Case Report

A 30-year-old woman was seen for evaluation of secondary infertility. A past pregnancy ended in artificial abortion when she was 20 years old. Menstruation began at age 11. She complained of severe dysmenorrhea and moderate dyspareunia. Pelvic examination revealed a retroverted uterus that was regular in contour but swollen to approximately 11 weeks' gestational size. Transvaginal ultrasound revealed thickening of the myometrium and diffuse foci distributed throughout the myometrium.

Investigation of the patient's infertility began in 1991 at

a private hospital. The results of hysterosalpingography suggested deformity of the uterine cavity and left tubal occlusion. The right tubal patency was normal. GnRH-a was administered from September 1991 to March 1992 for clinically suspected endometriosis. The patient was referred to our hospital in August 1994 for further examination and adjuvant therapy. An infertility screening test was conducted, but there were no abnormal findings other than an enlarged uterus and elevated tumor marker levels. MR images suggested diffuse adenomyosis involving the entire uterine myometrium (Figure 1). The abnormal tumor marker levels indicated CA-125 in the luteal phase : 77 U/ml (cutoff<35 U/ml).

In September 1994, a laparoscopic study revealed stage II endometriosis (r-AFS score (1985): 2 points), and adenomyosis was suspected because the uterus was swollen to 12 weeks' gestational size, and there were deep lesions in the subserosal myometrium. Blue berry spots were also seen in the vesico-uterine peritoneum (Figure 2). A needle biopsy was taken from the uterine myometrium. Vasopressin (Pitressin injection; Sankyo Co. Ltd., Tokyo, Japan) was dissolved in 4 ml of saline solution and injected in the sub-serosa of the uterus. Then a biopsy was taken with a single needle puncture instrument (SR-26178 DH; Karl Storz Co. Ltd., Germany) from the uterine myometrium through the sub-serosa. The biopsy revealed endometrial glands and stroma tissue within the myometrium, confirming the diagnosis of adenomyosis (Figure 3).

Despite the severe symptoms, complication of mild endometriosis and adenomyosis, because of the patient's wish to remain fertile, we decided to avoid hysterectomy. The treatment selected was expectant therapy, i.e., electrocautery of the endometriotic implants, laparoscopic uterosacral nerve ablation and irrigation of the peritoneal cavity. Two months after expectant therapy, the patient conceived without medical treatment. Unfortunately, a spontaneous abortion occurred after 18 weeks of gestation.

Address Correspondence : Dr. Akira Fujishita, Department of Obstetrics and Gynecology, Nagasaki University School of Medicine, 1-7-1 Sakamoto, Nagasaki 852-8501, Japan



Figure 1 Sagittal T-2 weighted MR image (SE 600/30) of the pelvis shows the uterus markedly enlarged and high intensity spots in the myometrium. The junctional zone in both sections is unclear. (a) SE 600/30 and (b) SE 2,000/80.



Figure 3 Endometrial glands and stroma tissue within the myometrium. The arrowhead shows the uterine serosa. (H & E, final magnification, $\times 33$)

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Figure 2 (a) Adenomyosis in a uterus as seen through laparoscopy and (b) blue berry spot (arrow) in the pelvic peritoneum.

Discussion

Hysterectomy is usually the therapy of choice for adenomyosis. Recently, preoperative diagnosis of adenomyosis is made by vaginal ultrasound or laparoscopic surgery⁵⁾. Percutaneous or transcervical myometrial biopsy is a possible alternative for patients with adenomyosis, especially those who wish to remain fertile. However, according to another report, sensitivity is low and diagnosis rate depends on the number of biopsies and the depth of adenomyosis⁶⁾. Improvements in myometrial biopsy will undoubtedly lead to a standard method of treatment. Our successful diagnosis of adenomyosis by needle biopsy may be attributable to our having coincidental observed endometrial foci in the subserosal myometrium.

Nowadays, various ultrasonographic and radiological studies are being described for the diagnosis of adenomyosis. Bohlman et alⁿ, suggested that ultrasound may aid diagnosis by showing that a uterus is moderately enlarged and has an anechoic, thickened posterior portion. Fedele et alⁿ, also reported that diagnostic capability can Akira Fujishita et al : Laparoscopic Diagnosis of Adenomyosis

be enhanced by transvaginal ultrasonography. The sensitivity of transvaginal ultrasound findings was 80%, and the specificity was 74%, showing that ultrasound was a quick, non-invasive and useful method for the diagnosis of diffuse adenomyosis. Mark et al⁹⁾. studied 21 patients with clinically suspected adenomyosis using MRI and correctly predicted all eight cases in which the disease was confirmed histologically. Togashi et al¹⁰⁾. also performed a prospective study to evaluate the ability of MRI to differentiate between adenomyosis and uterine myoma. MRI accurately diagnosed the cause of uterine enlargement in 92 of 93 cases. Only one diagnosis of adenomyosis was mistaken. However, microscopic studies failed to explain the definitive difference in intensity between areas of adenomyosis and myometrium. Our case revealed typical adenomyosis findings by MRI and diffuse adenomyosis by ultrasonography, then confirmed the diagnosis using a laparoscopic needle biopsy taken from the uterine myometrium.

Adenomyosis is often accompanied by additional pelvic pathology such as uterine leiomyoma. It also occurs concomitantly in 6-20% of patients with endometriosis¹¹. Our case of adenomyosis was also complicated with pelvic endometriosis. GnRH-a therapy has demonstrated efficacy in reducing the size of uterine fibroid and in suppressing the extent and activity of endometriotic implants. There are four other reported cases of successful treatment of the symptoms of severe adenomyosis with GnRH-a¹⁻⁴⁾. GnRH-a therapy was successfully used to produce a hypoestrogenic environment in endometriosis and uterine myoma. However, there were side effects, such as abnormal lipid metabolism, bone loss, and certain symptoms in the hypoestrogenic state. Our patient had previously received GnRH-a therapy and had complained of hot flashes and moderate headache. Fedele et al¹². reported that there was no marked difference between GnRH-a therapy and expectant management in the treatment of infertility patients with minimal or mild endometriosis. In view of the possible side effects and the patient's refusal of GnRH-a therapy, we decided upon expectant management. It is still unclear how fertility is promoted by GnRH-a therapy, but our patient conceived two months later without medical treatment.

Only three cases of pregnancy confirmed after GnRH-a treatment of adenomyosis have been reported, with one pregnancy ending in a spontaneous abortion after 10 weeks of gestation³⁾. Another report described an early viable

pregnancy, but the outcome of the pregnancy was not published²). Only one case was a normal delivery⁴). Another patient did not conceive¹. Our patient conceived soon after laparoscopic treatment, but spontaneous abortion occurred after 18 weeks of gestation. The relationship between the patient's spontaneous abortion and adenomyosis is unknown. Further studies are needed to determine the pathogenic mechanism of infertility in women with adenomyosis. Whether the adenomyosis treatment selected is GnRH-a therapy, reduction surgery or expectant therapy, the physician must take into account the uterine size, fertility period and other factors of infertility.

Finally, in accordance with the trend toward minimally invasive surgery, laparoscopy is gaining importance in gynecologic surgery. A laparoscopic approach for the diagnosis of adenomyosis may have advantages over other surgical procedures. Because this is the first case of adenomyosis diagnosed by laparoscopic needle biopsy technique, we recommend further studies to develop a standard a pre-operative diagnosis of the disease.

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