

Monochorionic twin delivery after conservative surgical treatment of a patient with severe diffuse uterine adenomyosis without uterine rupture

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A 31-year-old nulliparous woman with severe diffuse uterine adenomyosis, which replaced nearly the whole uterine myometrium, visited our hospital due to severe dysmenorrhea, menorrhagia, and a desire to have a baby. The patient had a history of two spontaneous abortions. Laparotomic adenomyomectomy with transient occlusion of uterine arteries (TOUA) was performed safely and the patient tried *in vitro* fertilization and achieved an intrauterine twin pregnancy after recovery time of the operation. At 31+6 weeks of gestation, a male neonate baby weighing 1,620 g and a male neonate baby weighing 1,480 g were born by transverse lower segment cesarean delivery. There was no complication after the operation. The babies were discharged after receiving routine neonatal intensive care for neonatal respiratory distress syndrome. Adenomyomectomy with TOUA technique would be an option for conservative surgical treatment in patients with severe diffuse whole uterine adenomyosis. This is the first report of twin pregnancy after diffuse whole uterine adenomyomectomy with TOUA.

Keywords: Adenomyomectomy; Adenomyosis; Transient occlusion of uterine arteries; Twin

Introduction

Adenomyosis, is characterized by heterotopic endometrial glands and stroma in the myometrium, and is an established cause of menorrhagia, dysmenorrhea and pelvic pressure symptoms [1]. Uterine adenomyosis is becoming more common with an increasing trend of pregnancy at a later age. Also, there has been an increase in the incidence of severe diffuse uterine adenomyosis, which invaded the whole uterus in young reproductive-aged women who desire to have a baby. We have previously reported transient occlusion of uterine arteries (TOUA) laparotomic adenomyomectomy for diffuse adenomyosis in two studies [2,3] for relief of symptoms and future pregnancy. However, there is no data to assess the feasibility and safety of diverse pregnancies in women who undergo operative treatment for severe diffuse whole uterine adenomyosis.

A few studies have introduced diverse surgical techniques for uterine-sparing surgical treatment for uterine adenomyosis, which was almost characterized as focal but not specific

diffuse whole uterine adenomyosis. Also, there is a lack of case reports or statistical analysis of ongoing twin pregnancy outcome following diffuse adenomyomectomy. In this literature, we want to introduce a course of conception and delivery of monochorionic twins after extensive resection with diffuse adenomyomectomy using TOUA.

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Case report

A 31-year-old nulliparous woman was referred to our clinic for the treatment of severe diffuse adenomyosis. The patient had severe dysmenorrhea, dyspareunia and lower abdominal pain and she had a history of two early spontaneous abortions, although she was trying to get pregnant for the last two years. Previously, she had used progestin and gonadotropin releas-

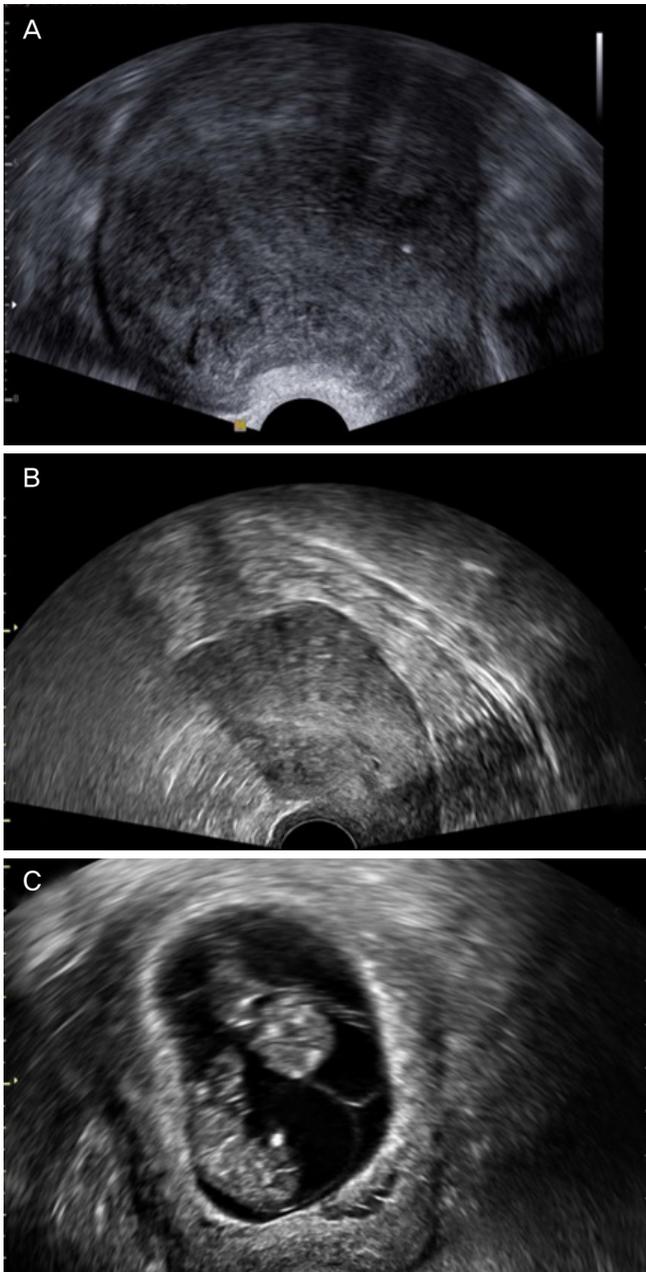


Fig. 1. Transvaginal ultrasound sonogram show severe diffuse uterine adenomyosis (A). Down-sized uterus after adenomyomectomy (B) and fetal sonogram at 11 weeks (C) .

ing hormone agonist to relieve the symptoms of adenomyosis medically, but they had no effect. A transvaginal ultrasound showed diffusely invaded and whole uterine adenomyosis (Fig. 1A). The patient wanted to receive conservative surgical treatment for relief of symptoms and future pregnancy. TOUA laparotomic adenomyomectomy was performed safely.

The procedure of TOUA laparotomic adenomyomectomy was performed in the same manner as in our published report [2]. The operation time was 80 minutes and the estimated blood loss was 400 mL without any intraoperative complications. Final pathology showed uterine adenomyosis. As a post-operative management option, gonadotropin releasing hormone agonist (leuprolide acetate 3.75 mg) was consecutively administered three times for three months. After 20 months later, she visited our clinic for ultrasonographic examinaion to plan for a baby. She had anteverted, normal sized uterus and the endometrium was clear (Fig.1B).

Due to personal reasons, the woman tried to get pregnant 30 months later. *In vitro* fertilization was successfully performed and an intrauterine monochorionic twin pregnancy was confirmed (Fig. 1C).

At 21+0 weeks of gestation, she was admitted for careful monitoring of uterine contractions and any other expected complications. During antenatal care, the myometrium thickness was monitored regularly, and there was no defect in the thickness of the myometrium on ultrasound. At 25 weeks, there was continuous uterine contraction with shortening of cervix. For lung maturation of fetuses, betamethasone 12 mg was injected intramuscularly, twice at 24 hours interval. At 31+6 weeks of gestation, we decided to perform a caesarean delivery to prevent future abrupt fatal complications, which included uterine rupture, monochorionic pregnancy complications in the 3rd trimester. Two male neonates weighing 1,620 and 1,480 g were delivered by caesarean section. There was no gross finding of uterine rupture but the peritoneum and bladder were adherent to the uterus; hence, bladder injury had to be repaired after delivering the baby. The myometrium and bladder were sutured separately and uterine contractions were sufficient and there was no atony or bleeding during the postpartum period. One week later, there was no leakage of urine on cystography and the Foley catheter was removed. The mother was discharged without any other problems and the babies were discharged after three months of prematurity care, without any postnatal complications.

Discussion

Adenomyosis is usually divided into diffuse and localized adenomyosis, according to the extent of the disease. Especially, there is a few report of diffuse uterine adenomyomectomy. Unlike the focal type, diffuse adenomyosis is associated with difficulties in surgical treatment to maximize the amount of excised adenomyosis and to improve the myometrial integrity. There have been a few studies to overcome the limitation of surgical management such as severe intra-operative bleeding [2,4,5]. We have previously reported two studies and the aim of our technique is to reduce the operative complications by achieving intraoperative bleeding control with TOUA. The efficacy and results of TOUA have been described in the published reports [2,3].

Although there were no adverse outcomes such as uterine rupture or postpartum hemorrhage in this case, there was bladder injury due to severe peritoneal adhesion. Reducing adhesion formation is another challenge for future fertility and safe delivery.

A suspected cause of uterine rupture after uterine surgery is excessive use of electrocauterization, which induces necrosis of the myometrium and disturbs scar healing. Intra-operative

bleeding control by TOUA can reduce the frequency of use of electrocautery during the operation, which can help in uterine wall regeneration.

To the best of our knowledge, this report presents the first case of twin delivery without uterine rupture. All of the reported cases led us to be cautious about uterine rupture during subsequent pregnancy after adenomyomectomy. Meticulous surgical technique is important to minimize the uterine defect. Also to prevent uterine rupture, admission and close monitoring of uterine contraction with regular checking of uterine wall thickness should be needed to make a decision of proper delivery time. Otsubo et al. [6] reported that uterine wall thickness is one of the risk factor of uterine rupture after diffuse adenomyomectomy.

A uterine rupture is one of catastrophic obstetrical complication after uterine surgery including adenomyomectomy. Including latest published report, there were eight cases of uterine rupture after adenomyomectomy [6-10]. Six of cases are laparoscopic surgery and the other two cases are laparotomic surgery (Table 1) [6-12]. The gestational age of the rupture typically occurs around the 30th weeks of gestation. In case of twin pregnancy after laparoscopic adenomyomectomy the uterus was ruptured with sudden onset of severe abdomi-

Table 1. Reported case of uterine rupture after adenomyomectomy

Reference	Modes of conception	Fetal number	Modes of surgery	Interval between operation and conception (mo)	Gestational age at the time of uterine rupture (wk)
Wood (1998) [8]	Spontaneous conception	Singleton	Laparoscopic myometrial electrocoagulation with excision of adenomyotic area	NA	12
Suginami et al. (2001) [11]	NA	Singleton	Laparoscopic adenomyomectomy	NA (>3)	32
Wada et al. (2006) [7]	IVF	Twin	Laparoscopic adenomyomectomy	12	30
Morimatsu et al. (2007) [9]	Spontaneous conception	Singleton	Laparoscopic adenomyomectomy	1	28
Onishi et al. (2011) [12]	NA	Singleton	Laparoscopic adenomyomectomy	NA (about 48)	31
Otsubo et al. (2016) [6]	IVF	Singleton	Laparotomic adenomyomectomy	22	19
Otsubo et al. (2016) [6]	IVF	Singleton	Laparotomic adenomyomectomy	36	16
Nagao et al. (2015) [10]		Singleton	Laparotomic adenomyomectomy	14	35

NA, not available; IVF, *in vitro* fertilization.

nal pain. It was unexpected event just 10-hours after using tocolytics [8].

In this case, it was inevitable to make decision of preterm delivery to prevent uterine rupture while monitoring of uterine contraction and uterine wall thickness. The patient had contraction with short cervix measured of 1cm at 31wks of gestation. Despite more than 50% of singleton pregnancies after surgery of diffuse uterine adenomyosis have delivered normally via elective cesarean section [11], there was only one report of twin pregnancy after laparoscopic adenomyomectomy and it was ruptured at 30wks of gestation. We made decision of preterm delivery but it was an effort to prevent uterine rupture. If we had waited until full-term, the risk of uterine rupture might be increased. In case of pregnant women with premature uterine contraction who had adenomyomectomy before pregnancy, admission and close monitoring is recommended to make a decision of adequate time to deliver.

To be on the safe side, in women who have undergone diffuse adenomyomectomy, single-embryo transfer might be recommended [8], but our current report suggests that twin pregnancy is not an absolute contraindication for in vitro fertilization treatment, which can demonstrate that multiple pregnancy without a severe adverse outcome is possible after adenomyomectomy. But further study and more number of cases are needed to establish the safety and efficacy of this management method.

In our department, there were 5 cases of full term cesarean deliveries in women who underwent diffuse adenomyomectomy with TOUA method, all of which were cases of singleton pregnancies without uterine rupture. These data could support technical safety and efficacy of TOUA adenomyomectomy. A previously reported study enrolled 26 patients with severe diffuse adenomyosis, and recently, 6 patients among these twenty-six patients were pregnant and one patient had twin pregnancy. TOUA adenomyomectomy is an effective method of conservative surgery for diffuse uterine adenomyosis [2].

In conclusion, adenomyomectomy with TOUA technique would be an option for conservative surgical treatment in patients with severe diffuse whole uterine adenomyosis. Because adenomyomectomy has been performed very recently, there is no consensus guideline of antenatal care of pregnant women who had diffuse adenomyomectomy. To be safe delivery, obstetricians have to be cautious about uterine rupture. Close

observation of uterine contraction and sonographic evaluation should be needed to a pregnant women who had adenomyomectomy.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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