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Case Report

Uterine rupture at 33rd week of gestation after laparoscopic myomectomy with signs of fetal distress. A case report and review of literature

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ABSTRACT

Objective: We describe a case of uterine rupture (UR) during pregnancy after laparoscopic myomectomy (LM) and discuss the risk factors of UR.
Case report: A 37-year-old woman with multiple myomas underwent laparoscopic myomectomy. Subserosal and intramural myomas were enucleated, and the myometrial wounds were repaired with single-layer suturing. Sixteen months after the operation, the patient conceived. At 33 weeks of gestation, emergency cesarean section was performed for the indication of fetal distress. A male neonate was delivered without asphyxia. During cesarean section, surgeons identified a 2 × 3 cm myometrial defect at one of the myomectomy sites, and diagnosed incomplete UR. The myometrial defect was repaired with debridement and suturing.
Conclusion: Based on the literature review, the risk of UR during pregnancy after LM is estimated to be less than 1% when all the surgical procedures have been performed appropriately. Myomectomy should be performed with careful consideration by surgeons who have good knowledge of the wound healing process in the myometrium.

Introduction

Laparoscopic myomectomy (LM) has become a common surgical procedure for treating symptomatic leiomyoma. LM is a preferable alternative to abdominal myomectomy (AM); it clearly reduces postoperative pain, shortens hospital stay, allows a quicker return to normal activity, and helps prevent postoperative adhesions. However, it has been suggested that LM is associated with longer operative times and increased risk of uterine rupture (UR) during subsequent pregnancies.

Although UR during pregnancy is rare, it can be a catastrophic obstetric complication associated with a high rate of maternal and fetal morbidity and mortality [1]. The most important risk factor affecting UR during pregnancy is a uterine scar created by previous uterine surgery such as cesarean section, myomectomy, adenomyomectomy, hysteroscopic resection, or surgery to treat ectopic pregnancy, but other factors have also been reported, such as congenital uterine anomalies, abnormal placentation, and induction of labor. This case report was exempt from the institutional review board at our institute.

Case

A 37-year-old primigravida was referred to our hospital because she had a uterine myoma (10 cm in diameter) with symptoms of dysmenorrhea and hypermenorrhea. Transvaginal ultrasonography and magnetic resonance imaging (MRI) revealed 4 uterine myomas: a subserosal myoma (10 cm in diameter) with a narrow stalk originating from the uterine fundus, a subserosal myoma (4 cm in diameter) originating from the center of the anterior uterine body, an intramural myoma (3 cm in diameter) located on the right side of the anterior uterine body, and an intramural myoma (1 cm in diameter) located in the center of the posterior uterine body (Fig. 1). For preoperative management, the patient received 4 months of subcutaneous gonadotropin-releasing hormone analogue therapy (Leuplin, Takeda, Tokyo, Japan, 1.88 μg), and the size of all myomas were decreased (8.5 cm, 3 cm, 2.5 cm, 0.8 cm in diameter respectively) at the time of operation.

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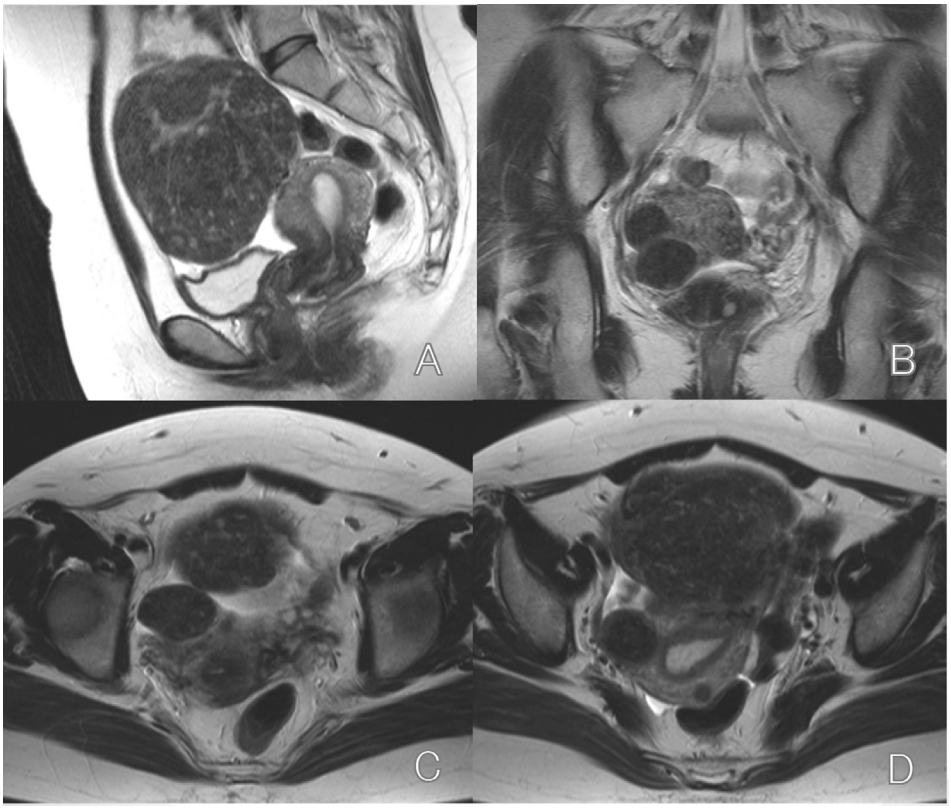

Fig. 1. Magnetic resonance imaging (MRI) of uterine myomas. MRI revealed 4 uterine myomas: 1) a subserosal myoma (10 cm in diameter) arising from the uterine fundus (A), 2) a subserosal myoma (3.5 cm in diameter) arising from the center of the anterior wall (B, C), 3) an intramural myoma (2.5 cm in diameter) located on the right side of the anterior wall (B, D), and 4) a small intramural myoma in the posterior uterine body (D).

Intraoperatively, the 2 subserosal myomas were enucleated by cutting their stalks with an ultrasonic scalpel (Harmonic Ace, Johnson & Johnson, New Jersey, USA) (Fig. 2A–D). Next, the two intramural myomas were enucleated by incising the uterine muscle layers above the myomas longitudinally with monopolar electrocautery. They were removed with gentle traction and rotational force by laparoscopic forceps with assistance from the ultrasonic scalpel inserted into the space between the myoma and the uterine muscle layer. All incisions in the myometrium were repaired with single-layered z-sutures with 0 Vicryl (Johnson & Johnson, New Jersey, USA) (Fig. 2E–I).

Sixteen months after the operation, the patient conceived spontaneously. At 32 weeks gestation, she was admitted to our hospital with a diagnosis of threatened premature labor. She had symptoms of frequent uterine contraction, irregular abdominal pain, and shortened cervical length of 10 mm. Cardiotocographic monitoring (CTG) revealed a reassuring fetal status pattern (Fig. 3A). No abnormal findings were identified with fetal ultrasonography, including amniotic fluid volume and placentation. Uterine contractions were well controlled after admission with continuous intravenous administration of ritodrine hydrochloride. Approximately 6 days after admission, CTG revealed occasional variable decelerations with less variability. At 33 weeks gestation, CTG revealed loss of variability with frequent variable decelerations (Fig. 3B) and decreased amniotic fluid volume (AFI, 2 cm). We decided to transport the patient to another hospital for further management of the pregnancy and intensive care for the neonate. Due to prolonged fetal bradycardia just before transport (Fig. 3C), emergency cesarean section was performed immediately after arrival at the other hospital. A male neonate weighting 1679 g was delivered. He appeared non-asphyxiated, and his Apgar scores at 1 and 5 min were 8 and 9, respectively. Umbilical arterial gas analysis indicated no acidosis (pH, 7.318) and respiratory distress syndrome did not occur. After closure of the uterine cesarean incision, the surgeon became aware of a myometrial defect 2 × 3 cm in size that reached the endometrium on the anterior uterine wall near the right uterine horn (Fig. 4A and B). He diagnosed incomplete UR at the site of a previous LM scar. Debridement of the lesion and 2-layer myometrial suturing was performed (Fig. 4C). The pathological diagnosis of the removed myometrial specimen was focal necrosis of myometrium. The mother and neonate had an uneventful puerperal and neonatal course, respectively.

Discussion

Uterine leiomyomas are the most common benign pelvic tumors in women of reproductive age. Although they are often asymptomatic, sometimes they cause menorrhagia, dysmenorrhea, and pelvic pressure. They might also impair fertility through several mechanisms [2]. For patients who wish to preserve fertility, myomectomy is the preferred surgical procedure over hysterectomy, although some other uterus-preserving approaches are available, such as uterine artery embolization, magnetic resonance-guided focused ultrasound, and medical treatment including GnRH analogues or sex steroids [2,3].

LM is a recently introduced surgical technique used to treat uterine myomas. LM is an excellent alternative to AM. Compared with AM, LM is clearly associated with less blood loss, reduced postoperative pain, shorter hospital stay, and quicker return to normal activity. Pregnancy rates and spontaneous abortion rates are comparable to AM. LM also prevents postoperative adhesions, which is particularly advantageous when pregnancy is desired [2,4,5].

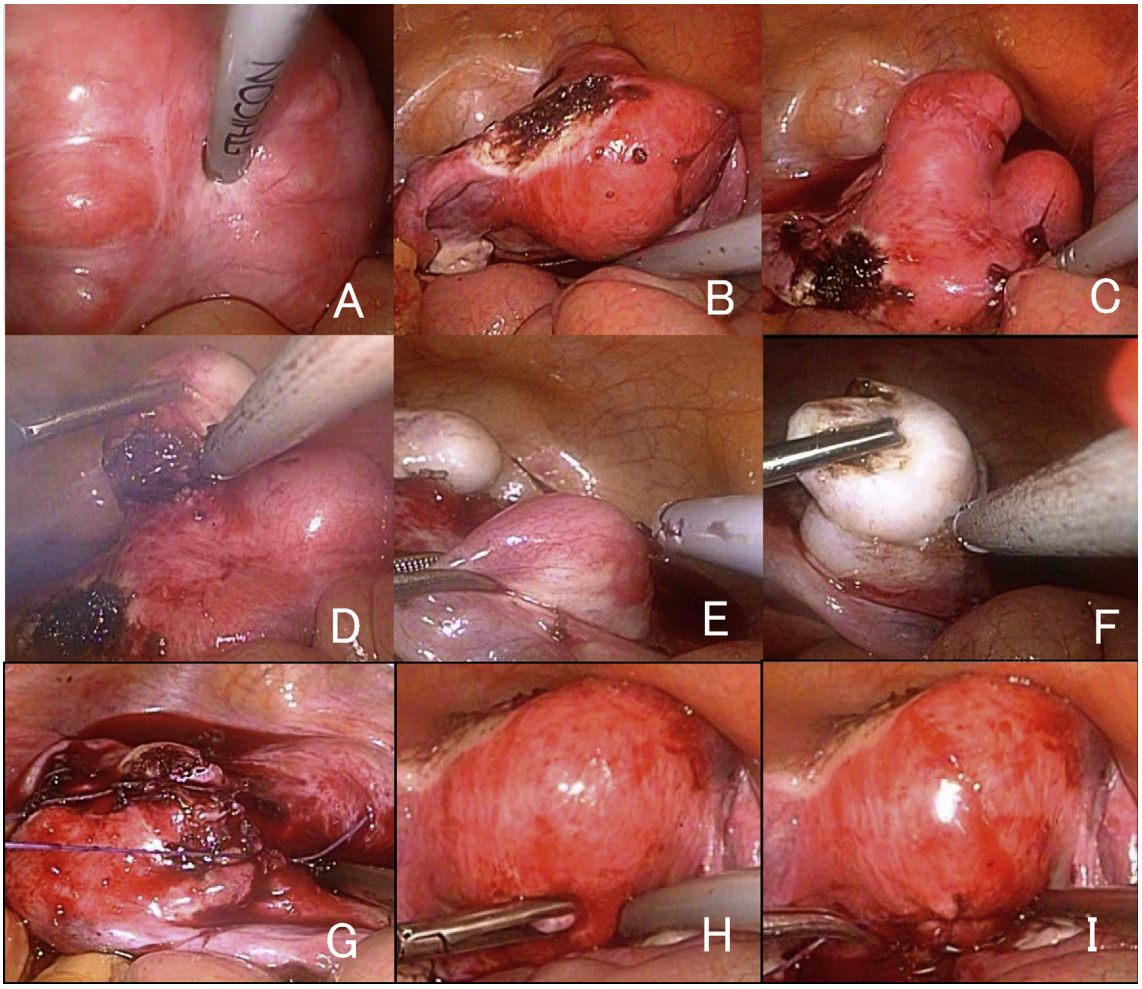

Fig. 2. Laparoscopic myomectomy. The subserosal myoma in the fundus was removed by cutting its narrow stalk (A, B). Another subserosal myoma, on the anterior wall, was removed by cutting its stalk (C, D). The intramural myoma on the anterior wall was enucleated (E, F). All of these incisions were repaired using single-layer z-sutures (G). The small myoma on the posterior wall was enucleated and the incision was repaired using the same suturing method (H,I).

One of the major concerns regarding myomectomy in women of reproductive age is the risk of UR during subsequent pregnancy or labor. Although UR during pregnancy is a very rare occurrence, it is a catastrophic obstetric complication that is associated with high maternal and fetal morbidity and mortality. The most important risk factor associated with UR in pregnancy is uterine scarring caused by previous cesarean section [6] or other uterine surgeries, such as myomectomy [2–4], adenomyomectomy [7,8], hysteroscopic procedures [9], or surgery to treat ectopic pregnancy [10].

Incidence of uterine rupture during pregnancy after laparoscopic myomectomy

Although there are many reports concerning the relationship between UR and previous cesarean section, it is difficult to determine the incidence of UR after previous uterine surgery. Dubuisson et al. reported a detailed analysis of UR after LM in 2000 [4]. Among the 100 patients who delivered after LM, there was only 1 spontaneous UR along the LM scar, so the authors concluded that the rate of UR after LM was 1.0%. Some other studies have also reported the incidence of UR during pregnancy after LM. Sizzi et al. prospectively studied 2050 women who underwent LM. Out of 309 deliveries, UR occurred in 1 woman (0.26%) [11]. Koo et al. reported that out of 523 patients who had follow-up through the end of pregnancy, 3 URs occurred (0.6%) [12]. Kim et al. reported that out of 54 pregnancies after LM, no UR occurred but 1 patient had a uterine wall defect identified during elective cesarean section (uterine dehiscence rate, 1.8%) [13]. Tian et al. reported that out of 81 pregnancies, 4 uterine scar defects that required suture repair were identified during elective cesarean section, although no UR occurred (uterine dehiscence rate, 4.9%) [14]. Although Bernardi et al. reported a very high incidence of UR (10%; 4 URs in 39 deliveries) [15], many researchers have reported a significant number of pregnancies or deliveries (over 500 cases in total) without any URs after LM [2,3,16]. It is difficult to determine the actual incidence of UR during pregnancy after LM, as some reports of UR do not describe the incidence of procedures or pregnancies and deliveries, especially case reports [2]. According to the large clinical trials, case reports, and review articles mentioned above, the incidence of UR during pregnancy after LM is estimated to be no higher than 1% when the myometrial incision is appropriately repaired [2,3,16].

It is unclear whether there is a greater risk of UR after LM than AM. The incidence of UR after AM is estimated to be relatively low because many researchers have reported a significant number of pregnancies without UR or only a few cases of UR after AM [3,4,13,14]. However, Roopnarinesingh et al. reported a high UR rate (5.3%) after AM in 1985 [17]. On the other hand, the incidence of UR after laparoscopic or laparotomic adenomyomectomy seems to be

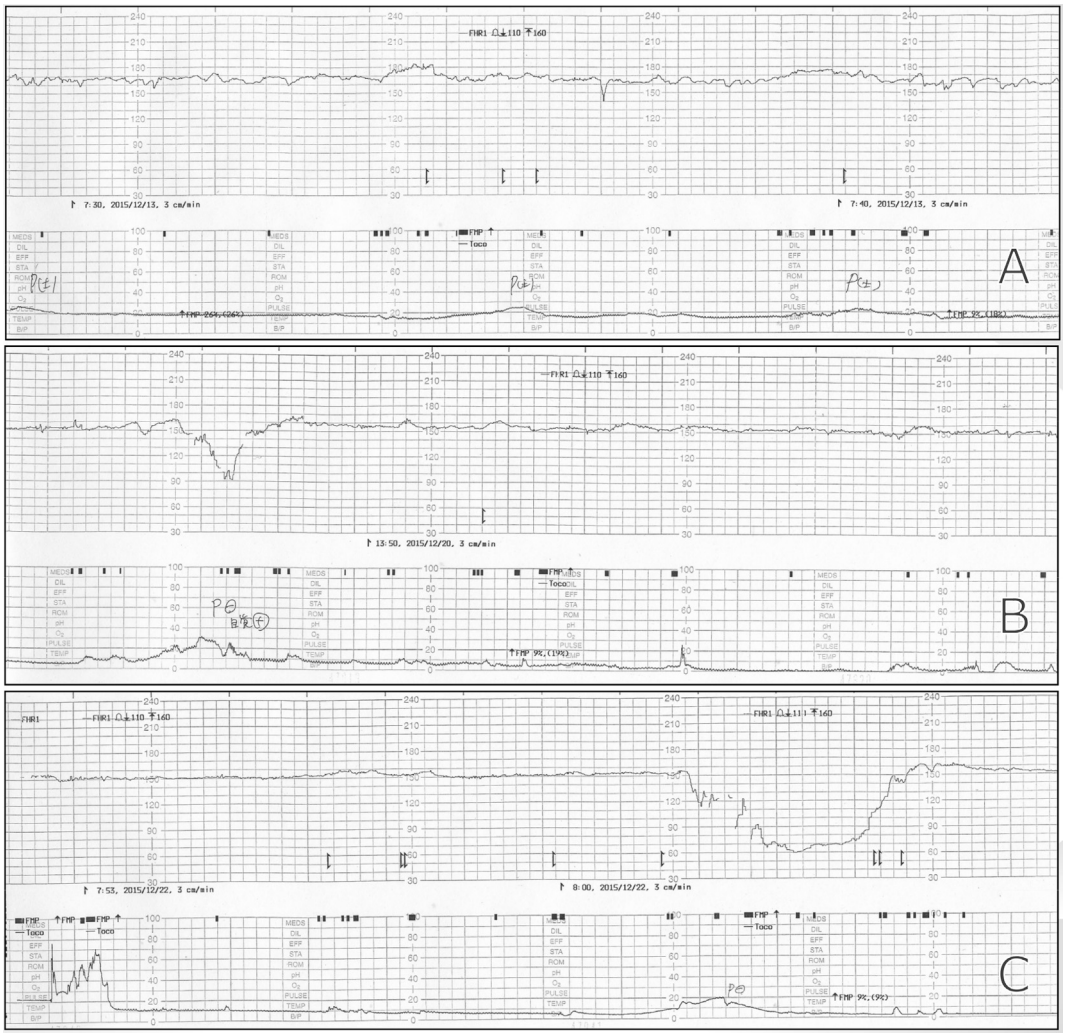

Fig. 3. Cardiotocographic monitoring (CTG). At administration (32 weeks gestation), CTG revealed a reassuring fetal status pattern with frequent uterine contraction (A). At 33 weeks gestation, CTG revealed loss of variability with frequent variable deceleration (B). Prolonged fetal bradycardia occurred just before transporting the mother to another hospital (C).

higher (6.3%–8.7%) than that after myomectomy [7,8], although we found fewer reports on pregnancy after adenomyomectomy than after cesarean section or myomectomy.

Risk factors affecting uterine rupture after laparoscopic myomectomy

Many authors have discussed risk factors for UR or dehiscence during pregnancies after LM. In particular, two groups reviewed cases of UR subsequent to LM (19 and 7 cases, respectively) to determine whether common causal factors could be identified [16,18]. Both review articles identified that the most important risk factors for UR are related to surgical technique, which could affect wound healing, rather than myoma characteristics. They proposed that excessive use of electrocautery for hemostasis should be avoided because it results in poor vascularization and can induce necrosis of the myometrium, which would lead to impaired scar healing and decreased tensile strength in the myometrium [4,9,14]. Expeditious suturing of the myometrium is preferable to electrocautery for achieving hemostasis, even in LM [18]. Suturing technique was found to be more important for wound healing than the number of suture layers. Multi-layer closure should be used to repair myometrial defects for deep and intrusive myomas, although some reports indicated that a 2-layer suture closure is not superior to 1-layer myometrial closure [19]. Single-layer suture should be performed even for superficial myomectomy, because there have been reports of UR in women who have undergone subserosal LM [4,20]. Excessive suturing can induce a foreign body reaction and tissue ischemia that interferes with proper muscle healing and reproductive function [18,21]. Adequate repair does not depend on the number of suture layers but rather on full-thickness, evenly placed suture placement, and avoiding hematoma formation [19].

In Table 1, we summarized details about surgery and obstetric outcomes in 33 reported cases of UR after LM [12,13,16,18]. In most cases, electrocautery was used for hemostasis and the uterine defect was either repaired with a single layer of sutures or sutures were not used. Cases where only sutures were used for hemostasis and more than 2-layer of sutures were used for repair were significantly less common in these reported cases of UR, although the numbers of patients with each factor at baseline were unknown. It should be noted that there were 20 reported cases of UR after subserosal LM. When we evaluated the 33 cases of UR in more detail, we did not find significant differences in myoma characteristics (intramural: 36.3% vs subserosal ∓ pedunclated: 60.6%), myoma size (≤3 cm: 30.2% vs >3 cm: 60.6%), use of electrocautery for hemostasis (monopolar: 25.8% vs bipolar: 32.3% vs

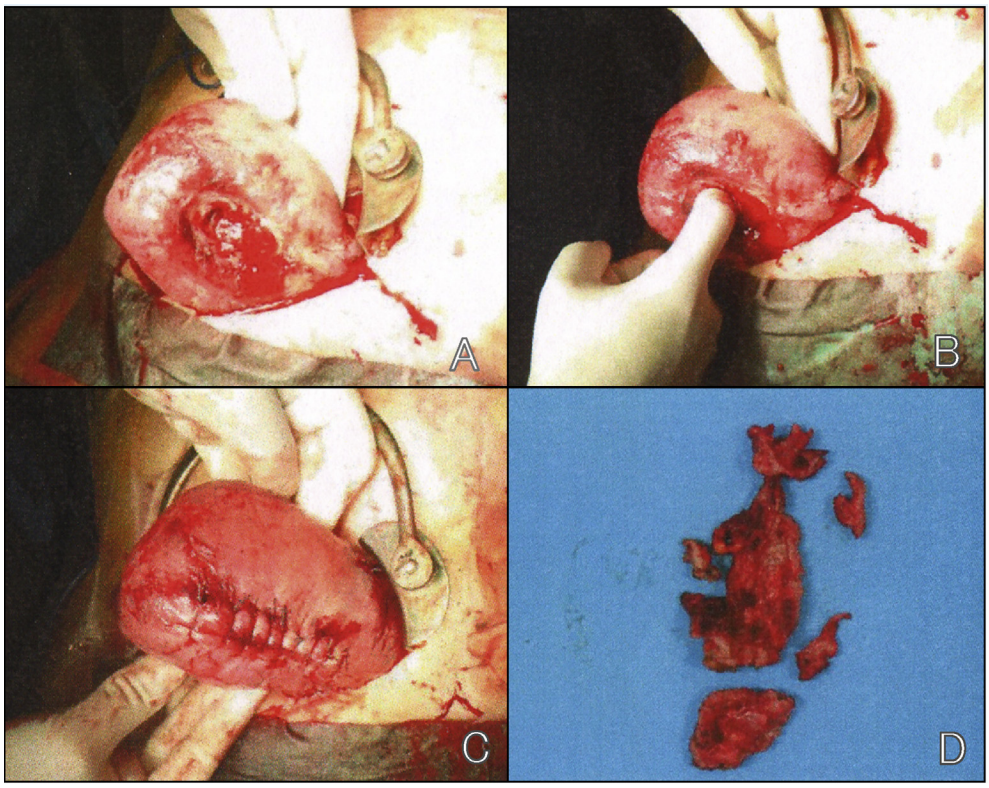

Fig. 4. Uterine rupture identified during cesarean section. The myometrial defect reached the endometrial cavity. It was 2 × 3 cm in size and located on the anterior uterine wall near the right uterine horn (A, B). Debridement and 2-layer myometrial suturing was performed (C). Macroscopic picture of resected tissues in ruptured site of myometrium (D). The pathological diagnosis of the removed myometrial specimen was focal necrosis of the myometrium.

bipolar + suture: 35.5%), and number of suture layers (no suture: 33.3% vs 1-layer: 50.0%) between the groups compared.

Gambaccorti-Passerini et al. also recently reviewed UR after myomectomy [22]. In their 2016 review, they selected 11 reliable articles and evaluated detailed data about trial of labor and related pregnancy outcomes after myomectomy. A total of 1034 pregnancies and 745 deliveries at >24-weeks were included in the review. They identified 7 cases of UR. The incidence of UR prior to the labor (1.52%, 5/330) was higher than that in the group who had a trial labor (0.47%, 2/426). The incidence of UR was 0.99% (6/606) after LM and 0.67% (1/150) after AM. When they compared suturing techniques and myoma characteristics of ruptured cases, and the use of electrocautery for hemostasis and myoma locations between UR group and non-UR group, no statistically significant differences were observed. They concluded that a trial of labor after myomectomy (TOLAM) could be as relatively safe as a trial of labor after cesarean delivery (TOLAC). Thus, TOLAM might be considered a feasible and possible safe option. Although UR after myomectomy is a complication that is difficult to predict because it can happen at any time during pregnancy, evaluation with ultrasonography or even MRI may be useful to identify women at higher risk of UR during pregnancy after myomectomy.

Regarding other risk factors, individual characteristics related to healing such as growth factor production or excess collagen deposition [23], carbon dioxide pneumoperitoneum [24], and surgical skill of the surgeon [4] may affect wound healing. The authors of the review articles concluded that LM should be performed by adequately trained and experienced surgeons that take all precautions to minimize the risk of UR, including limited use of electrocautery for hemostasis and adequate closure of the myometrium [16,18].

Some authors focused on the importance of saving the “myoma pseudocapsule” to enhance wound healing as part of the LM procedure to avoid complications [21,25,26]. Tinelli et al. demonstrated that laparoscopic intracapsular myomectomy (LIM) preserves the myoma pseudocapsule, which contains two important neuropeptides (neuropeptide substance P and vasoactive intestinal peptide) that possibly enhance the normal healing of the uterine scar and myometrial function in subsequent pregnancies [18,25,26]. They proposed that LIM should always be performed to maximize the potential for future fertility and minimize the risk of UR during pregnancy [25].

Adequate interval of contraception after laparoscopic myomectomy

An interval of contraception after myomectomy to ensure adequate wound healing might be necessary. Some authors have evaluated changes in uterine structure (resolution of hematoma, absorption of suture materials, decrease in scar size, etc.) during the recovery process after myomectomy using MRI [27], ultrasonography [28], and 3-D power Doppler ultrasonography [29]. These 3 papers all concluded that the wound healing process seems to be almost complete by 3 months. Since the appropriate interval of contraception may also depend on the number, location, and depth of enucleated myomas, the surgeon should decide on the interval necessary for each patient. On the other hand, because there are some reports of myomectomy during pregnancy and subsequent deliveries without any UR or dehiscence, prolonging the interval of contraception alone may not reduce the risk of UR and such an interval is not a definitive risk factor of UR [14,30,31]. According to the evidence above about the duration of wound healing, we recognize that at least 3 months is needed and we recommend 6 months of contraception to patients as a precaution. It is also very important to obtain informed consent before myomectomy about the need for an adequate interval of contraception after the procedure.

Table 1
Surgical details and obstetric outcomes of reported cases of uterine rupture after laparoscopic myomectomy.

Case No.	Myoma type	Size (cm)	EM entry	Hemostasis	Suture	UR gestation (weeks)	Fetel survival	Maternal survival
1	IM	ND	Yes	Endocoagulator	1-layer	28	ND	Yes
2	SS	3	No	MP	1-layer	34	Yes	Yes
3	IM	3	No	BP + suture	1-layer	34	Yes	Yes
4	IM	5	Yes	BP + suture	1-layer	28	Yes	Yes
5	IM	5	Yes	suture	ND	28	Yes	Yes
6	IM	ND	Yes	BP	2-layer	29	Yes	Yes
7	IM	9	No	BP + suture	2-layer	33	Yes	Yes
8	SS	5	No	BP	No	33	No	Yes
9	SS-P	11	No	MP	No	34	Yes	Yes
10	SS	ND	No	MP	No	17	No	Yes
11	ND	2.5	No	suture	3-layer	28	No	Yes
12	SS	8	No	BP	No	40	Yes	Yes
13	SS-P	1,2, 1.2	No	MP	No	29	Yes	Yes
14	SS	2	No	BP	1-layer	33	Yes	Yes
15	SS	2	No	BP	1-layer	33	Yes	Yes
16	SS-P	4	No	BP	No	35	Yes	Yes
17	SS-P	4	No	MP	No	36	Yes	Yes
18	IM	2.4	No	MP	1-layer	36	Yes	Yes
19	IM	4	No	BP	1-layer	35	Yes	Yes
20	SS	5	No	BP + suture	2-layer	35	Yes	Yes
21	SS	4	No	BP + suture	1-layer	34	Yes	Yes
22	SS	3	No	BP	No	34	Yes	Yes
23	SS	5	No	BP + suture	1-layer	38	Yes	Yes
24	SS	8	No	BP + suture	1-layer	24	No	Yes
25	SS	6	No	BP + suture	1-layer	35	Yes	Yes
26	SS	2.5, 2, 2	No	BP	No	36	Yes	Yes
27	IM	5	No	BP + suture	2-layer	37	Yes	Yes
28	SS	5	No	BP + suture	1-layer	32	Yes	Yes
29	SS	7	No	BP + suture	1-layer	21	No	Yes
30	IM	4	No	BP	1-layer	35	Yes	Yes
31	IM	4	No	MP	ND	17	No	Yes
32	IM	4.5	ND	ND	ND	27	No	Yes
33	SS-P	2	No	MP	No	22	No	Yes

IM = intramural myoma, SS = subserosal myoma, SS-P = subserosal pedunclated myoma, BP = bipolar, MP = monopolar, ND = no data, [reference]: Case No. 1–19 [16], 20–26 [18], 27–29 [12], 30–33 [13].

Signs of uterine rupture

The majority of URs after LM occur before the start of labor, especially in early third trimester [13,16,18], although most URs after cesarean section occur during labor [6]. The most common early signs of UR or dehiscence during pregnancy are vaginal bleeding, increased uterine contraction, pain, discomfort, and reduction of fetal movement. Patients should be informed that they must immediately report any of these suspicious symptoms during pregnancy [3,16]. Signs of fetal distress on CTG sometimes occur before UR, as in our case, although there are case reports of UR occurring without any signs of fetal distress [5]. It is very important for the management of post-LM patients that all precautions should be taken to avoid missing the signs of UR or dehiscence during pregnancy.

Case evaluation based on above discussion

We evaluated our patient's case based on the above discussion. In the patient's previous LM procedure, the intramural myoma of the ruptured site was clearly removed with preservation of its pseudocapusule (Fig. 2E–F). Electrocautery was not used for hemostasis since no bleeding was detected during enucleation; instead, a 1-layer suture was used. It is possible that the 1-layer suture was insufficient for wound repair because the myoma was deeply intruded into the myometrium even though it was a small nodule (Fig. 1C). We could not rule out this might have led to hematoma formation and induce the necrosis of myometrium.

Although it is unclear whether the UR was directly related to the cause of fetal distress, we estimated that necrosis of the pregnant myometrium could induce chronic uterine contraction leading to decreased amniotic fluid volume, fetal distress, and delivery of a small-for-date infant. Earlier detection of the signs of fetal distress may have led to the delivery of a well-being neonate.

Conclusion

LM is an excellent alternative for women who desire uterine preservation, because it is minimally invasive and associated with fewer perioperative complications than AM. The risk of UR during pregnancy after LM is estimated to be less than 1% when all the surgical procedures are performed appropriately. Myomectomy should be performed by well-trained and experienced surgeons in selected patients after careful consideration of the wound healing process in the myometrium. This includes techniques such as enucleation with preservation of the myoma pseudocapsule, hemostasis without excessive coagulation using electrocautery, and adequate suturing to avoid hematoma formation, especially in the laparoscopic procedure.

Conflict of interest

None of the authors have any conflicts of interest to declare.

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