Uterine adenomyosis and adenomyoma: the surgical approach

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The appropriate surgical treatment of adenomyosis, a benign invasion/infiltration of endometrial glands within the underlying myometrium, remains a subject of discussion. Since 1990, in place of the classical V-shaped resection method, various kinds of surgical management have been attempted, including a uterine muscle flap method that emphasizes fertility preservation, an asymmetric dissection method, and various modified reduction methods. Laparoscopic adenomyomectomy has also become an alternative to laparotomy for surgically managing the focal type of adenomyosis, although it seems to be associated with a higher risk of uterine rupture than laparotomy. This article reviews the surgical treatment of adenomyosis, including 23 uterine rupture cases that occurred during post-adenomyomectomy pregnancies, and provides an updated picture of the state of the field. (Fertil Steril® 2018;109:406–17.

Key Words: Adenomyosis, adenomyomectomy, uterine rupture, wound healing, Triple-flap method

Adenomyosis is a benign gynecologic tumor and is classified into diffuse or focal adenomyosis, depending on the extent and location of the disease as well as the histological classification (1, 2). The surgical treatment of adenomyosis remains a subject of discussion. Since 1990, in place of the classical V-shaped resection method (3, 4), various forms of surgical management have been attempted, including a uterine muscle flap method that emphasizes fertility preservation (5–8), an asymmetric dissection method (9), and various modified reduction methods (10, 11). Laparoscopic adenomyomectomy has also been attempted as an alternative to laparotomy for surgically managing the focal type of adenomyosis (12–15), although it may be associated with a higher risk of uterine rupture than laparotomy. As many reports pertaining to the surgical treatment for adenomyosis have been published (16–19), this article mainly reviews the post-1990 literature, focusing on the 23 uterine rupture cases that occurred during post-adenomyomectomy pregnancies.

A search of the main medical literature shows that 2,365 cases of adenomyomectomy have been reported since 1990, including 2,123 (89.8%) in Japan. A total of 397 post-procedural pregnancies were reported, with 337 (84.89%) resulting in live births and 23 instances of ruptured uterus. Adenomyomectomy is a recognized treatment for certain types of dysmenorrhea and menorrhagia. However, in postoperative pregnancies, it is characterized by a high miscarriage rate, thinning of the uterine walls, and silent uterine ruptures occurring during the mid-term pregnancy. A higher incidence of placenta accretes and placenta percreta compared to cesarean section and myomectomy has also been reported (15). Electrically-powered instruments are associated with most cases of uterine rupture, and a causal relationship is suspected (13, 20, 21).

INDICATIONS

Adenomyomectomy is now at the stage where new surgical methods are being tried, but the indications for surgery differ depending on the surgeon. The indications include dysmenorrhea and hypermenorrhea that are difficult to control with medication, infertility and recurrent miscarriages, and a desire to preserve fertility or the uterus. Focal adenomyosis resection is performed using either laparotomy or laparoscopic surgery, whereas diffuse adenomyosis is limited to using laparotomy. Laparoscopic procedure can be used for treating focal adenomyosis, although it entails a risk of leaving some of the lesions unexcised. Laparoscopy is also used in conjunction with laparotomy for treating diffuse adenomyosis, to reduce operative stress such as adhesion. For the treatment of diffuse adenomyosis, however, since the size of the lesion and of the areas involved

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and the boundary between the lesion and the normal tissues can only be grasped by palpation, laparotomy or preferably laparoscopy-assisted laparotomy is required in order to excise the lesion accurately and to repair the uterine wall by suturing in layers.

PREOPERATIVE EXAMINATIONS

Magnetic resonance imaging (MRI) examination is performed in order to grasp accurately the location and extent of the uterine adenomyosis and the position of the uterine cavity in order to determine the site, direction, and depth of the incision to be made into the uterus. Hysterosalpingography is also performed to examine the shape and size of the uterine cavity (5, 8, 20).

SURGICAL TREATMENT FOR ADENOMYOSIS

The conservative surgical treatment for adenomyosis in young women was first reported in 1952 (4). Subsequently, the partial excision of an adenomyosis, as a cytoreductive surgery, became common after the introduction of wedge resections, in which the uterine wall is excised in a V-shape.

Prior to the 1970s, the most common suturing materials were silk and catgut (both plain and chromic). These heterogeneous protein materials contributed to complications, such as suture failure, due to strong responses to foreign materials. Nevertheless, adenomyomectomies continue to be performed due to the development of absorbable sutures that elicit less severe tissue responses, as well as to the development of powered devices, such as electric, ultrasonic, high-frequency scalpels, which have resulted in surgeries with minimal bleeding.

LAPAROTOMIC SURGERY: PARTIAL REDUCTION SURGERIES

Wedge Resection of the Uterine Wall

In this classic technique, parts of the serosa and uterine adenomyoma are removed via wedge resection, after the part of the seromuscular layer where the adenomyoma is located has been identified using laparotomic or laparoscopic methods. In this procedure, part of the adenomyoma tissue may remain on one or both sides of the incision. The uterine wall wound created by the adenomyoma resection is sutured together with the remaining muscular layer and serosa (22). The postoperative clinical effectiveness on dysmenorrhea and menorrhagia is small, and recurrence occurs due to the presence of remaining adenomyomatous tissue.

Modified Reduction Surgery

These include various laparotomic and laparoscopic technique modifications (11, 13, 14, 20, 23–36). In 1991, a modification of the partial adenomyosis excision was reported to have been performed on 37 patients. This involved cutting the adenomyomatous tissue into thin slices using a microsurgical technique in conjunction with laparotomic surgery. Consequently, 6 women become pregnant, and none miscarried (23).

In 1993, another laparotomic modification involving partial excision of adenomyoma was reported to have been performed on 28 patients. Of these, 18 attempted to conceive, with 13 achieving clinical pregnancies. Eventually, there were 9 (50.5%) live births and 7 (38.8%) miscarriages (24).

Transverse H Incision of the Uterine Wall

There has been a report describing a laparotomic modification which compared 5 women treated with the classical method with 6 women undergoing modified reduction surgeries involving the transverse H incision technique (10). The transverse incision was made on the uterine fundus, using an electro-surgical scalpel, separating the uterine serosa from the uterine myometrium. After widely opening the bilateral uterine serosa, the adenomyoma tissue was removed using an electro-surgical scalpel or scissors. A tension-less suturing technique was used to apposition the myometrial edges and close the wound in one or two layers. The first layer of sutures was applied to close the defect in the uterine wall and establish hemostasis. The bilateral serosal flaps resulting from the vertical incision, and the upper and lower flaps resulting from the transverse incision, were closed with a sub-serosal interrupted suture.

In the later study by the same author, based on data collected up to 2010, out of the 41 patients who underwent the H-incision technique, 31 attempted to conceive; and 12 (38.7%) achieved clinical pregnancy, 5 (16.1%) miscarried and 7 (22.5%) reported live births (25). One other study reported on 14 women who underwent this technique (26). All of them wished to conceive; and 3 (21.4%) achieved pregnancy and all had healthy babies.

Wedge-shaped Uterine Wall Removal

This method has been reported in a recent study where adenomyoma is resected with a thin margin (wedge-shaped removal), after a sagittal incision in the uterine body. The radical resection involves the laminate layers on both the endometrial and serosal sides. Reconstruction of the uterine wall involves suturing using the continuous horizontal mattress technique. The external serous layer is sutured such that the cut edges are inverted to reduce incisional adhesion to the omentum, intestines, and peritoneum. This suturing technique involves the 'baseball' or continuous Lembert stitch method. Among 103 patients, 70 attempted to conceive during the study period, of them, 21 (30%) achieved clinical pregnancies; and 16 (22.8%) achieved live births (27).

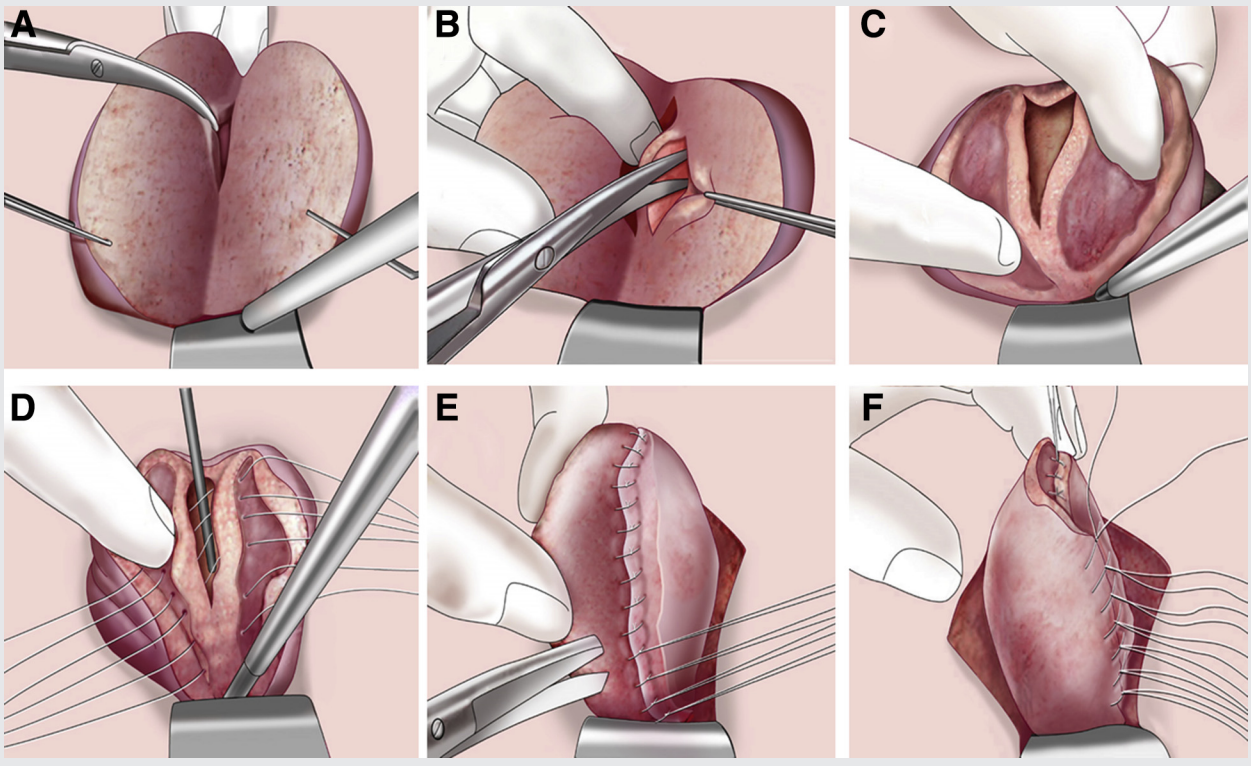
LAPAROTOMIC SURGERY: COMPLETE ADENOMYOSIS EXCISION

Triple-flap Method

This adenomyomectomy technique is based on a completely new idea that differs from conventional surgical methods. The method involves reconstructing the uterine wall defect using normal uterine muscle. The technique is not only effective for diffuse uterine adenomyosis, but also for nodular adenomyosis, and it has the potential to contribute to the prevention of uterine ruptures during postoperative pregnancies (5–8,20).

The technique has the following three characteristics: complete extraction of the uterine adenomyosis (Fig. 1A

FIGURE 1



Example of Triple-flap method applied to the posterior uterine wall adenomyosis (Adapted from Osada H. Shikyusenkinsho. [Uterine adenomyosis.] In: Osada H. Jissen fujinka fukkukyoka-shujutsu. [Laparoscopy for gynecology: a comprehensive manual and procedure DVD]. Tokyo: Medical View, 2009 [8].).

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and B and Fig. 2A and B) by performing adenomyomectomy, followed by wound repair to avoid suture failure and wound re-opening using a cold knife (non-electrical scalpel); reconstruction of a uterine cavity which can sustain subsequent pregnancy (Fig. 1B and C and Fig. 2B and C), in which an endometrial uterine muscle flap is prepared by metroplasty through opening the uterine cavity and removing the uterine adenomyosis under palpation; and reconstruction of a uterine wall resistant to rupture during a subsequent pregnancy (Fig. 1D–1F and Fig. 2D–2F). The uterine muscle on the serosal side is used to fill the large uterine wall muscle defect.

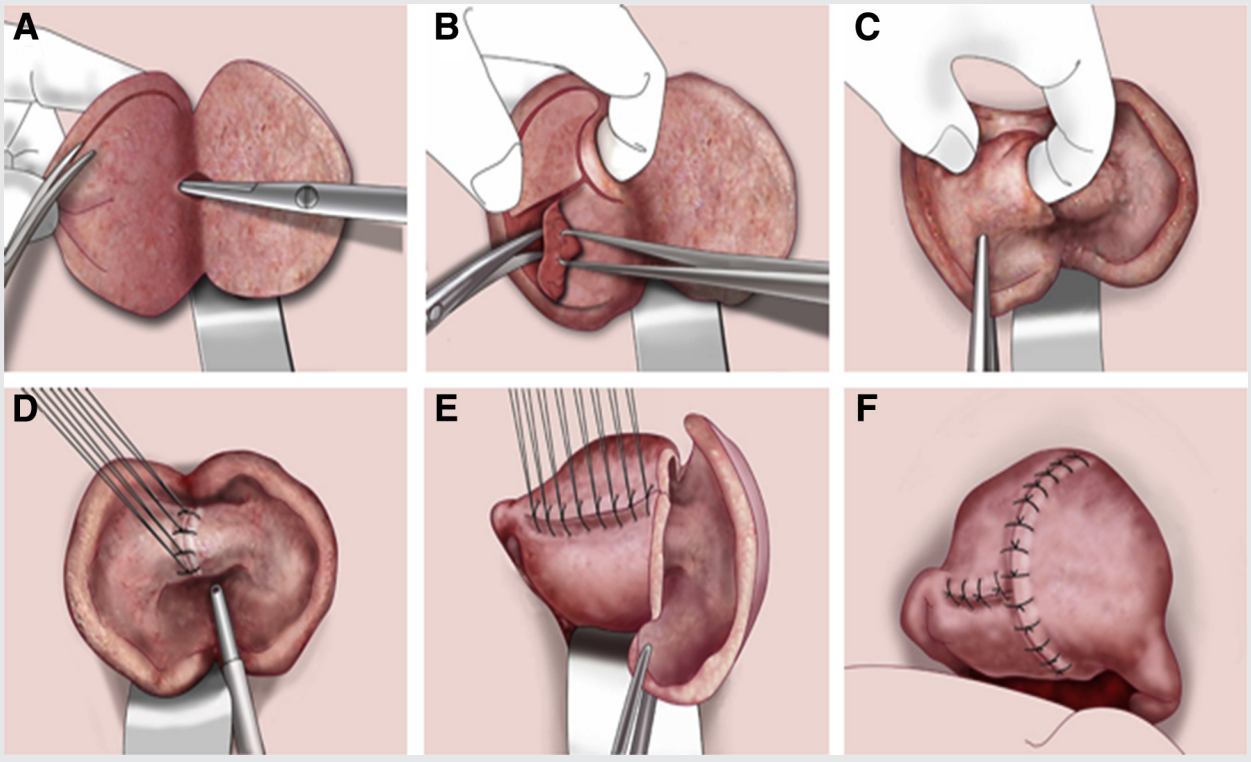
A study (20) which examined 113 women who underwent this method demonstrated that the blood flow in the operated area had returned to normal in almost all cases (92/113, 81.4%) within 6 months. Of 62 women who wished to conceive, 46 conceived and 32 delivered a healthy baby by elective Cesarean section. There was no case of uterine rupture. During the 27 years of the study period, only 4 cases (3.5%) relapsed and required surgical treatment. In cases where a uterine adenomyosis resection is performed without opening the uterine cavity and the uterine wall is formed by superimposing a uterine muscle flap from the serosal side, the procedure is referred to as the double-flap method (12, 13).

As this surgical procedure is extremely difficult to perform by laparoscopic surgery only, because the extirpation of the adenomyoma should be carried out under palpation and delicate suturing by hand is required, open surgery is preferable. However, as long as extirpation and suturing are performed under simple open surgery, laparoscopically-assisted adenomyomectomy can also be performed (20, 28).

Asymmetric Dissection Method

This procedure involves asymmetric dissection of the uterus longitudinally, using a round-type loop electrode and a high-frequency cutter, followed by retracting the uterine fundus upwards using a silk suture, and then cutting the uterine adenomyoma into slices. From the incision, the myometrium is dissected diagonally as if hollowing out the uterine cavity. It is followed by a transverse incision to open the uterine cavity (Fig. 3B). While inserting the index finger into the uterine cavity, the adenomyosis lesion is excised to >5 mm of the inner myometrium (Fig. 3C). The lesion is then excised to >5 mm of the serosal myometrium on the left uterine side (Fig. 3D). Afterwards, the uterine cavity is sutured and closed, followed by uterine reconstruction, with the left side covering the right side (Fig. 3E). The serosa is continuously sutured, using the same suture to rejoin the uterus (Fig. 3F). To date, 1,349 patients have undergone this technique (9). Post-operative spontaneous uterine rupture was seen in 5 cases in this series (29).

FIGURE 2



Example of Triple-flap method applied to the anterior and posterior uterine wall adenomyosis (Adapted from Osada H. Shikyusenkinsho. [Uterine adenomyosis.] In: Osada H. Jissen fujinka fukkukyoka-shujutsu. [Laparoscopy for gynecology: a comprehensive manual and procedure DVD]. Tokyo: Medical View, 2009 [8].).

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LAPAROSCOPIC ADENOMYOSIS SURGERIES

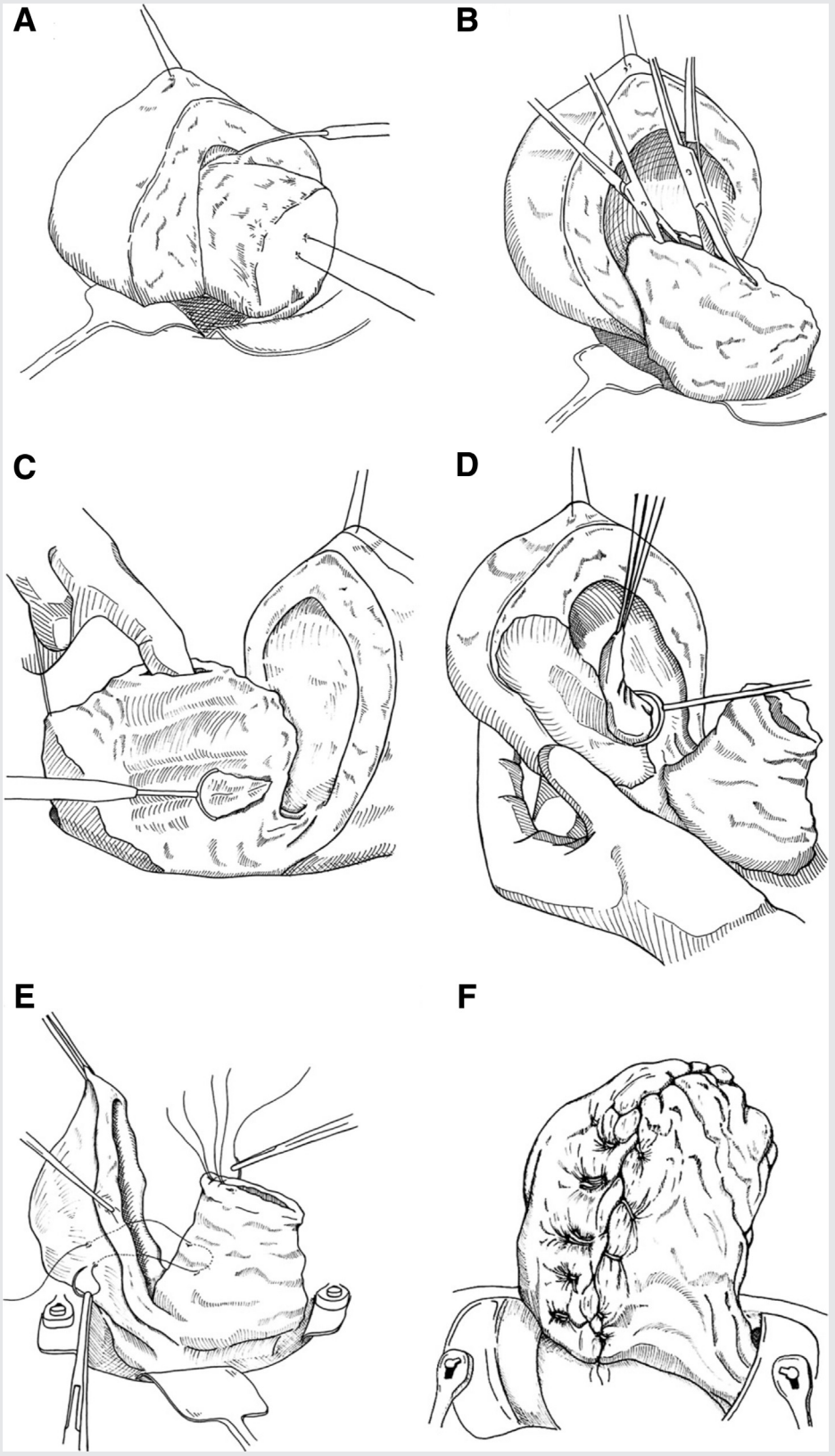
With the laparoscopic approach, the surgical methods are limited due to the surgery allowing only limited directions of movement and limited instrumentation use; further, palpation is impossible. For nodular types of adenomyosis, laparoscopic surgery is possible using a surgical approach that is similar to that used for uterine fibroids (11–15,24,30–32). However, diffuse-type lesions require extensive resection and complicated suturing, necessitating difficult operations involving advanced techniques.

The laparoscopic surgical method includes a longitudinal (37) or transverse (12, 13) incision of the uterine wall along the adenomyoma. This is followed by resection of the adenomatosis using a monopolar needle (30) or a laser knife (11, 14). Then suturing of the endometrial cavity, if perforated, and suturing of the uterine wall are performed (which may be accomplished in the seromuscular layer (33)), in two or more layers (38) or using the double-flap method (12, 13). Finally, removal of the adenomyotic mass may be accomplished using a morcellator (37) or removal bag. However, there is a concern that a morcellator may disperse fragments of undetected malignant tumors throughout the abdominal cavity and upstage otherwise contained cancer (39), and its use has been discouraged by the Food and Drug Administration since 2014. Some reports suggest that ultrasonic guidance is useful when the intraoperative recognition of the adenomatous lesion is difficult (33, 40).

In a case report by Morita et al. (41), cases of 3 women with focal adenomyosis who underwent lesion excisions involving procedures similar to those used for laparoscopic myomectomies are reported. There were no complications, and the patients were hospitalized for only 3 days. In the report, the authors concluded that accurate diagnoses and preoperative MRI evaluations of the adenomyosis are the most important indicators of successful surgery (41).

In the first report of laparoscopic adenomyomectomy using the flap method, the defective muscle resulting from the removal of a uterine focal adenomyosis, was reconstructed (12). In this procedure, 14 patients with focal adenomyosis ($\geq$30 mm in diameter), diagnosed using MRI, underwent adenomyosis extirpation that involved transverse incisions of the uterine bodies using monopolar needles. The flaps were overlapped and sutured to offset the lost muscle layer and reconstruct the uterus. In this procedure, if the uterine cavity is perforated, it is closed by suturing. In principle, however, this is a double method since it involves removal of the uterine adenomyosis without opening the uterine cavity. Postoperative pregnancies were achieved by 2 patients, and one had a vaginal delivery of a live infant. By 2017, this method had been performed by Kitade et al. (13) in 74 patients. Thirty-one patients desired pregnancy,

FIGURE 3



Example of asymmetric dissection method (**A**) Dissecting the uterus; (**B**) opening the uterine cavity; (**C**) excising the inner side lesion; (**D**) excising the outer side lesion; (**E**) suturing the lesion; and (**F**) rejoining the uterus. (Adapted from Nishida et al. Conservative surgical management for diffuse uterine adenomyosis. Fertil Steril 2010:94:715-9. [9].)

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and 13 (41.9%) were successful; 4 miscarried and 9 (29.0%) delivered live infants. There were no reported cases of uterine ruptures (13).

In a report of laparoscopic adenomyomectomy that included uterine rupture cases (14), 141 patients with focal adenomyosis underwent lesional excisions using laser knives. After the adenomyosis was removed, the uterine muscle layer was closed using continuous suturing (2-0 synthetic absorptive sutures) of the defect space. A total of 102 women desired to become pregnant, and the total clinical pregnancy rate was 31.4% (32/102). When the women were divided into age groups (those <40 years vs. those ≥ 40 years), the clinical pregnancy rates were 41.3% and 3.7%, respectively. In the older group, 5 of 6 pregnancies ended in miscarriages. Uterine ruptures were not observed in this series, but there were 2 cases of placenta accrete that resulted in postpartum hysterectomies.

Kodama et al. (30) reported cases of 71 patients with focal adenomyosis who underwent monopolar lesional excisions, which included uterine rupture cases. This method involved the convex lens method for lesion excision (15). Of the patients who underwent the procedure, 32 (45.1%) wished to conceive; 16 women achieved a clinical pregnancy, including 3 (18.7%) who miscarried; and 13 who delivered live infants (delivery rate, 40.6%). There was one case of uterine rupture.

POSTOPERATIVE PREGNANCY OUTCOMES

Post-adenomyomectomy improvements in dysmenorrhea and hypermenorrhea vary but are recognized. The postoperative pregnancy rate also varies between 17.5% and 72.7%. However, artificial reproductive technology largely contributes to the relatively high pregnancy rate. In total, 2,365 uterine adenomyomectomies have been reported from 18 facilities worldwide (Table 1) (11, 13, 14, 20–36). Of these, 2,123 procedures have been performed at 13 facilities in Japan, constituting 89.8% of the global total. Among these, 449 pregnancies have been confirmed and 363 (80.8%) resulted in deliveries including 2 cases of stillbirths. There were 13 (3.6%) cases of uterine ruptures (Table 1). An additional 11 cases of uterine rupture have been reported (Table 2).

UTERINE RUPTURE RISK

The frequency of uterine rupture in non-scarred uteri is 0.005% (42), but increases to 0.04%–0.02% in women with scarred uteri (43); vaginal births after cesarean sections further increase the risk to 0.27%–0.7% (44). Pregnancies occurring in women who have been treated for uterine adenomyosis have a higher risk of spontaneous rupture than do those without a history of surgery (45). In pregnancies following the surgical removal of uterine adenomyosis, the uterine rupture frequency is higher (20). A literature review suggested that the risk of uterine rupture after uterine adenomyosis is 6.0% (46). Regardless, the risk of uterine rupture due to pregnancy, after removal of a uterine adenomyosis, is $>1.0\%$ (47), compared to 0.26% (48) in pregnancies following myomectomy.

According to a 1986 review, there were only 29 cases of obstetric complications due to severe uterine adenomyosis over the preceding 80 years; thus, such complications were considered to be rare (49). I investigated the post-1990 literature for information regarding uterine ruptures due to pregnancy in women who had undergone the removal of uterine adenomyosis, and found a total of 24 cases reported from 18 centers. Further analysis suggested that these uterine ruptures occurred after adenomyomectomy involving laparotomic treatment in 13 cases, after laparoscopic surgery in 8 cases, and in other situations in 3 cases; 3 cases also resulted in hysterectomy due to uncontrolled bleeding. There were many cases in which uterine rupture was accompanied by complications, such as placenta accreta, placenta increta, and placenta percreta.

Uterine Rupture Due to Pregnancy after Laparotomic Adenomyomectomy

According to a 2008 report, Suginami et al. (11) performed laser adenomyomectomies in 138 cases (11). Of the 74 previously infertile women, 24 became pregnant, including 2 (8.3%) who experienced uterine ruptures. The course of the uterine ruptures is unknown. In 2016, Nishida et al. (29) reported 5 cases of uterine ruptures. To date, they have performed 1,349 cases of adenomyomectomy. Of the 176 patients who conceived, 221 postoperative pregnancies were confirmed, and uterine ruptures occurred in 5 women at 31, 27, 30, 16, and 19 weeks of gestation. The uterine rupture rate was 2.8% per case and 2.3% per pregnancy; in all cases, the uterine cavity was opened. The placenta had implanted in the sutured part of the endometrium in 5 cases, and 2 cases involved placental penetration. The symptoms of uterine rupture included abdominal pain without uterine contractions.

In 2014, Saremi et al. (27) reported the cases of adenomyomectomy using the wedge-shaped uterine wall removal technique on 103 women, including 57 infertile patients. Post-operative complications were observed in 6 patients, with 4 developing Asherman's syndrome and 2 experiencing spontaneous uterine ruptures (one at 37 gestational weeks and the other at 32 weeks). The 37-week pregnancy resulted in a stillbirth and the baby born at 32 weeks was transferred to a neonatal intensive care unit and survived. One patient had relapsed adenomyosis (27).

Uterine Rupture Due to Pregnancy after Laparoscopic Adenomyomectomy

The first report of uterine rupture due to pregnancy after a laparoscopic adenomyomectomy involved a twin pregnancy (50). Laparoscopic adenomyomectomy of focal adenomyosis was performed using monopolar cautery, and the residual myometrium was sutured with 1-0 poliglecaprone (25 sutures in two layers). The patient became pregnant 10 months after the adenomyomectomy, but her uterus ruptured after she experienced light abdominal pain at 30 weeks gestation followed by sudden abdominal pain during ritodrine infusion to prevent uterine rupture. Regardless, two male infants,

TABLE 1

Published cases of laparotomic and laparoscopic adenomyomectomy.

Author, year (ref.)	Patients	Operative method	Uterine incision	Patients who desired to conceive	Patients who conceived	Conceptions	Miscarriages	Deliveries	Uterine rupture
Kawamura et al., 1991 (23)	29	Laparotomic	Scissors	29	9	9	2 (22.2)	7 (24.1)	0
Kikuchi et al., 2004 (31)	24	Laparotomic	Monopolar	16	7	7[a]	2 (28.5)	3 (31.3)	0
Yosiki et al., 2004 (35)	67	Laparotomic	Monopolar	40	8	8	1 (12.5)	7 (17.5)	1 (12.5)
Suginami et al., 2008 (11)	138	Laparotomic, laparoscopic	Laser knife	74	24	NR	NR	24 (32.4)	2 (8.3)
Honda et al., 2009 (34)	51	Laparotomic	Metzenbaum, forceps	51	22	22	7 (31.8)	15 (29.4)	0
Fujishita et al., 2010 (25)	41	Laparotomic	High-frequency cutter	31	12	12	5 (41.6)	7 (22.5)	0
Nishimoto et al., 2011 (26)	14	Laparotomic	Scissors, monopolar, high-frequency cutter	14	3	3	0	3 (21.4)	0
Kishi et al., 2014 (14)	141	Laparoscopic	Laser knife	102	32	42	10 (23.8)	32 (31.3)	1 (3.2)[b]
Tanaka et al., 2014 (36)	11	Laparo. assist. adeno.	Monopolar	11	11	11	3 (18.7)	8 (72.7)	1 (12.5)
Kodama et al., 2015 (30)	71	Laparoscopic	Monopolar	32	14	16	3 (18.7)	13 (40.6)	1 (7.1)
Nishida et al., 2016 (29)	1349	Laparotomic	High-frequency cutter	NR	176	221	45(20.3)	176	5 (2.8)
Kitade et al., 2017 (13)	74	Laparoscopic	Monopolar	31	13	13	4 (30.7)	9 (29.0)	0
Osada et a1., 2017 (20)	113	Laparo. assist. adeno.	Metzenbaum, scalpel	62	32	46	14(30.4)	32 (51.6)	0
Fedele et al., 1993 (24)	28	Laparotomic	Monopolar	18	13	18	8 (44.4)[c]	10 (55.6)[d]	0
Grimbizis et al., 2008 (33)	6	Laparotomic	Scissors, monopolar	2	0	0	0	0	0
Kim et al., 2014 (28)	11	Laparo. assist. adeno.	Scalpel, Metzenbaum, monopolar	5	NR	NR	NR	0	0
Saremi et al., 2014 (27)	103	Laparotomic	Scalpel, monopolar	70	21	21	4 (19.0)	17 (24.3)[d]	2 (11.8)
Huang et al., 2015 (32)	94	Laparo. assist. adeno.	Scissors, monopolar	10	0	0	0	0	0
Total	2365			-	397	449	108	363	13 (3.6)

Notes: Data presented as n or n (%), unless stated otherwise. Studies with no record (NR) of data were not counted. Laparo. assist. adeno. = laparoscopy-assisted adenomyomectomy; Laparoscopic = laparoscopic surgery; Laparotomic = laparotomic surgery; Monopolar = monopolar cautery.
[a] Includes 2 cases of ongoing pregnancy.
[b] Case was not included in the researchers' 2014 study.
[c] Includes 1 ectopic pregnancy.
[d] Includes 1 stillbirth.

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weighing 1,585 g and 1,545 g, were delivered by emergency cesarean section, and both had 5-minute Apgar scores of 9. The ruptured posterior myometrium (7 cm long) was successfully repaired, after a blood loss of 2,600 ml. The mother's postoperative course was uneventful and the 2 infants continue to develop normally.

A second report (46) described a patient who became pregnant one month after undergoing a laparoscopic adenomyomectomy involving monopolar cautery. At gestational week 28, weak uterine contractions began, leading to intravenous ritodrine infusion. Severe abdominal pain and a non-reassuring fetal heart rate occurred abruptly, necessitating an emergency cesarean section. A 1,356 g baby was delivered, and the mother experienced a 2,560 ml blood loss. The uterine rupture, which had been reconstructed, occurred at the site of the previous uterine body surgery. The adenomyomectomy history and the short interval to pregnancy suggest the possibility of their contributing to an increased risk of uterine rupture.

In both of the above cases, the clinical courses had remarkable similarities. Both patients initially had weak uterine contractions during early pregnancy (30 and 28 weeks, respectively), ritodrine suppression was ineffective, uterine rupture occurred within short periods (2 and 10 hours, respectively), and the emergency cesarean sections and uterine reconstructions were successful.

In 2015, Kodama et al. (30) reported the outcomes of 71 laparoscopic adenomyomectomies that included one case of uterine rupture. Among the 71 patients, 32 attempted to conceive; 14 (43.8%) achieved clinical pregnancies once or

TABLE 2

Published cases of uterine ruptures after laparotomic and laparoscopic adenomyomectomy.

Author, year (ref.)	Patient age (y)	Operative method	Uterine incision	Contraceptive period (mo)	Modes of conception	Uterine rupture (wk)	Fetal number	Surgical treatment	Volume of bleeding (ml)	Fetal weight (g)	Fetal survival	Maternal survival
Yoshiki et al., 2004 (35)	NR	Laparotomic	Monopolar	NR	NR	26	Singleton	Preservation	NR	NR	Live	Live
Wada et al., 2006 (50)	33	Laparoscopic	Monopolar	12	IVF-ET	30	Twin	Preservation	2,600	1,585/1,545	Live	Live
Morimatsu et al., 2007 (46)	35	Laparoscopic	Monopolar	1	Spontaneous	28	Singleton	Preservation	2,560	1,356	Live	Live
Suginami et al., 2008 (11)	NR	NR	Laser knife	NR	NR	NR	Singleton	NR	NR	NR	NR	Live
Suginami et al., 2008 (11)	NR	NR	Laser knife	NR	NR	NR	Singleton	NR	NR	NR	NR	Live
Kasama et al., 2010 (51)	33	Laparotomic	Laser knife	36	IVF-ET	28	Singleton	Hysterectomy	6,130	1,274	Live	Live
Ukita et al., 2011 (52)	39	Laparotomic	NR	60	Spontaneous	29	Singleton	Hysterectomy	3,943[a]	1,614	Death	Live
Yazawa et al., 2011 (53)	37	Laparoscopic	NR	5	IVF-ET	33	Singleton	Preservation[b]	NR	1,956	Live	Live
Onishi et al., 2011 (54)	40	Laparotomic	NR	NR	IVF-ET	31	Singleton	Hysterectomy	5,200	1,700	Live	Live
Kishi et al., 2014 (14)	NR	Laparoscopic	Laser knife	NR	NR	NR	NR	NR	NR	NR	NR	NR
Tanaka et al., 2014 (36)	33	Laparo. assist. adeno.	Monopolar	36	IVF-ET	34	Singleton	Preservation	1,900	2,100	Live	Live
Kodama et al., 2015 (30)	41	Laparoscopic	Monopolar	4	Spontaneous	34	Singleton	Hysterectomy	5,150	2,032	Live	Live
Sato et al., 2015 (55)	35	Laparoscopic	NR	3	IVF-ET	28	Singleton	Preservation	NR	1,484	NR	Live
Nagao et al., 2016 (56)	42	Laparoscopic	NR	12	Spontaneous	35	Singleton	Preservation	NR	2,283	Live	Live
Nishida et al., 2016 (29)	38	Laparotomic	High-frequency	16	IVF-ET	31	Singleton	Hysterectomy	NR	NR	NR	Live
Nishida et al., 2017 (29)	35	Laparotomic	High-frequency	1	Spontaneous	27	Singleton	Preservation	NR	1,106	Live	Live
Nishida et al., 2018 (29)	31	Laparotomic	High-frequency	6	IVF-ET	30	Singleton	Preservation	NR	1,373	Live	Live
Nishida et al., 2019 (29)	34	Laparotomic	High-frequency	NR	Spontaneous	16	Singleton	Preservation	NR	NR	NR	Live
Nishida et al., 2020 (29)	32	Laparotomic	High-frequency	24	IVF-ET	19	Singleton	Preservation	NR	NR	NR	Live
Iwahashi et al., 2017 (57)	37	Laparotomic	High-frequency	72	IVF-ET	22	Singleton	Preservation	2140	NR	Death	Live
Yamaguchi et al., 2017 (58)	38	Laparotomic	NR	36	IVF-ET	33	Singleton	Preservation	759[a]	1,850	Live	Live
Wood et al., 1998 (59)	NR	Laparoscopic	Monopolar	24	NR	12	Singleton	NR	NR	NR	Death	Live
Saremi et al., 2014 (27)	NR	Laparotomic	Scalpel, monopolar	NR	NR	NR	Singleton	NR	NR	NR	NR	NR
Saremi et al., 2015 (27)	NR	Laparotomic	Scalpel, monopolar	NR	NR	NR	Singleton	NR	NR	NR	NR	NR

Note: High-frequency = high-frequency cutter; IVF-ET = in vitro fertilization-embryo transfer; Laparo. assist. adeno. = laparoscopy-assisted adenomyomectomy; Laparoscopic = laparoscopic surgery; Laparotomic = laparotomic surgery; Monopolar = monopolar cautery; NR = no record.

[a] Includes amniotic fluid.

[b] Adenomatoid tumor.

Osada. Uterine adenomyosis and adenomyoma. Fertil Steril 2018.

more, resulting in 3 (18.7%) miscarried cases and 13 (40.6%) live births. There was one case of uterine rupture. In this patient, the natural pregnancy occurred 4 months after the surgery. Despite normal health examination results, sudden abdominal pain occurred during gestational week 34. Thereafter, hemorrhagic shock occurred, necessitating an emergency delivery. During the laparotomy, a large amount of bloody ascites was observed, along with an 8 cm rupture site on the posterior wall of the uterine body. A hysterectomy was performed to control bleeding (intraoperative bleeding, 5,150 g), leading to a pathological diagnosis of placenta accreta or placenta increta. The 2,032 g newborn had a 2 minute Apgar score of 2 points and a 5 min score of 4.

There have been many other reports of uterine ruptures (51–56, 58–60). These reports point to post-laparoscopic adenomyomectomy pregnancies leading to uteri that may be easily ruptured by weak and short uterine contractions. These cases indicate that uterus ruptures are silent, and can be characterized as unpredictable.

FACTORS CAUSING UTERINE RUPTURES

The factors potentially related to uterine ruptures occurring during postoperative pregnancies include the adenomyosis removal method (e.g., cold knife, powered instruments), degree of extirpation of the adenomyosis (adenomyosis remnants in the tissue), extent and size of the uterine muscle defect, method of reconstructing the uterine cavity and the uterine wall, postoperative wound infection and postoperative hematoma formation, the period of contraception prior to postoperative pregnancy, and the skill of the surgeon.

WOUND HEALING DISTURBANCE DUE TO EXCESSIVE ELECTROCOAGULATION

Wound healing is generally a complex process involving inflammation, angiogenesis, new tissue formation, and tissue remodeling (61). This process requires balanced collagen deposition and growth factor release from the injured site, but pathological scarring (e.g., hypertrophic scarring) interferes with growth factor expression (62). Uterine ruptures involving the scarred area, after uterine fibroid nucleus surgery or Cesarean section, typically demonstrate abnormally high concentrations of collagen in the tissues near the rupture sites as well as fewer smooth muscle fibers. As a result, there is a high possibility that the strength of the uterine muscle layer has been undermined (61).

During laparotomic adenomyomectomies, the adenomyosis is removed using cold knives, such as scissors (25, 32, 33) Metzenbaums (20, 28, 34), scalpels (20, 27, 28), Kelly forceps (34), or powered instruments, such as monopolar electrocautery, bipolar electrocautery (27, 28, 30–33, 35–37, 49), high frequency cautery (25, 29, 62), and laser knife (11, 50). During laparoscopic adenomyomectomies, both unipolar motorized knives and harmonic scalpels are widely used (13, 36). The use of a powered instrument for removing the uterine adenomyosis seems to contribute to reduced blood losses and shortened operative times, contributing to an improved quality of life for the patient. During laparoscopic surgery in general, an electrocauterizer and/or laser, rather than cold instrumentation, are used. Although the vasopressin injection technique may be used in conjunction to reduce the hemorrhage during the procedure and the level of electrocauterization usage and there are some reports on its use for the treatment of ovarian endometriomas (63, 64), no study has been found on its use for adenomyomectomy. However, the use of powered instruments also results in hardening and discoloration of the incised tissue surface due to heat denaturation. With the increased prevalence of laparoscopic surgery since 1980, when powered instruments were introduced, these instruments have become necessary tools for making incisions and extracting tissue. During uterine adenomyopathy procedures, powered instruments are used in most facilities. As a result, there are more cases where tissue hardening and discoloration occur at the incision site, making the boundary between the abnormal and normal tissues less clear (20, 57).

On the other hand, a detailed histological investigation into the influence of electrocautery on wound healing has indicated that it may greatly influence wound healing and may cause secondary disability (65). Moreover, there is also a danger of wound dehiscence. Wound healing disorders may result in suture failures due to tissue necrosis, scarring, and excessive collagen deposition (66, 67).

Many papers have reported a histological delay in cure associated with electrocautery, compared with the use of a surgical blade (67–74). Hemostasis resulting from the use of a powered instrument causes the sealing of blood vessels due to the presence of clots of heat-denatured protein. At the same time, these clots form highly cohesive agglomerates due to tissue protein degeneration that affects wound healing (70).

CONTRACEPTION PERIOD PRIOR TO POSTOPERATIVE PREGNANCY

Whether a period of contraception after adenomyomectomy correlates with the occurrence of uterine rupture remains unclear. However, Bujold et al. (71) suggested that the interval between 1527 cesarean sections and the trial of labor after cesarean section correlated with the incidence of uterine ruptures; pregnancies occurring <24 months after a cesarean section had a 2- to 3-fold greater risk of uterine rupture. Following adenomyomectomy, the literature suggested that the contraception period varies by facility. Some facilities gave permission for pregnancies to be attempted within 3 months after surgery, but most facilities recommended contraception periods of 6–12 months. Interestingly, our literature review suggested that uterine ruptures, after adenomyomectomy, included 3 cases when the pregnancy occurred 3 months after surgery, 3 cases following 4–6 months of contraception, 2 cases after 7–12 months of contraception, and 4 cases after more than a year of contraception. Thus, it seems there is no certain correlation between the duration of the contraception period and the incidence of uterine ruptures (Table 2).

No reported investigation into the contraception period recommended prior to pregnancy, following surgical removal

of uterine adenomyosis, has been found. The postoperative field may be observed as an avascular area, when using colored Doppler imaging and contrast-enhanced MRI. The author has given permission for pregnancy to be attempted after confirming the resumption of blood flow (loss of the avascular area) to the postoperative field. In this area, blood flow resumed, in 92 cases (81.4%), within 6 months. However, when the uterine wall was largely resected, the resumption of blood flow could be delayed by >2 years (20).

LAPAROTOMY VERSUS LAPAROSCOPIC SURGERY

Currently there is no evidence indicating the best clinical and reproductive technologies for use in patients undergoing adenomyomectomies. Many investigators have described data in connection with the theoretical benefits of the principal techniques, but the results are based on relatively few cases, increasing the difficulty of showing statistically significant clinical differences. The published literature has also not elucidated the cause(s) of uterine ruptures.

Regarding the 23 uterine rupture cases after the removal of uterine adenomyosis, powered instruments (e.g., monopolar cautery, high-frequency cutter, laser knife) had been used in 15 cases and monopolar cautery and scalpel had been used in 2 cases. The remaining 6 cases had no description of the equipment used (Table 2). There were no reports of uterine ruptures resulting from surgeries involving the use of a surgical scalpel. Thus, it seems there is a definite association between uterine ruptures and the use of powered instruments.

Laparoscopic adenomyomectomy results in incompletely repaired muscle defects, compared with laparotomy surgery. Thus, the risk of uterine rupture is believed to increase during subsequent pregnancies following laparoscopic procedures (13, 20, 21, 72).

The efficacy of laparoscopy-assisted myomectomy has been reported by Nezhat et al. (73) in which the researchers found it to be a safe alternative to myomectomy by laparotomy. It is technically less difficult than laparoscopic myomectomy, allows better closure of the uterine defect, and may require less time to perform.

In the report published also by Nezhat et al. (74), the researchers found a large percentage of patients with symptomatic adenomas and adenomyosis have concurrent endometriosis. Overlooking the coexistence of endometriosis in women with symptomatic leiomyoma may lead to suboptimal treatment of fertility and persistent pelvic pain and it is important to be aware of the possibility of this association and to thoroughly evaluate the abdomen and pelvis for endometriosis at the time of myomectomy or hysterectomy in an effort to avoid the need for reoperation.

Adenomyoma is a hard tissue and when the lesion exists inside the myometrium, the boundary between the lesion and the normal tissue can only be grasped by palpation, which requires open surgery. Also due to its hardness, if left unexcised, the reconstruction of the uterine wall becomes difficult. Forced reconstruction of the uterine wall would strain the tissues abnormally and will likely result in ruptured sutures and anastomotic leakage. Therefore the diffused lesion must be thoroughly excised by laparotomy, preferably assisted by laparoscopy.

CONCLUSION

Uterine ruptures are directly linked to the endangerment of the lives of women and are a serious problem in perinatal management. Thus, in conjunction with the preoperative informed consent, providing information to patients and attending physicians regarding the importance of pregnancy management is vital. The fact that post-adenomyomectomy pregnancies have higher rates of uterine rupture than are associated with other surgical therapies has been clarified in this review. The frequency of these ruptures is much higher than that of those following myomectomies. However, the development of more reliable and safer surgical techniques, as well as the establishment of guidelines for pregnancies occurring after adenomyomectomy, will require further case accumulation and complication disclosures.

Nevertheless, based on this review, what emerged as the current best practice are as follows: Focal adenomyosis lesion can be treated laparoscopically. However, diffuse adenomyosis must be treated by laparotomy or preferably by laparoscopically-assisted laparotomy. Open surgery is safer than laparoscopy in that it can thoroughly excise the lesions to prevent the recurrence and to properly reconstruct the defect created by the surgery in order to prevent uterine rupture due to subsequent pregnancy. Although powered instruments seem to contribute to reduced blood losses and shortened operative times, such use also results in hardening and discoloration of the incised tissue surface due to heat denaturation, making the boundary between the abnormal and normal tissues less clear. No literature comparing the efficacy and/or safety of various cutting energies has been found. However, there are opinions suggesting that in general lasers cause least thermal denaturation. Although there is no consensus regarding the necessary contraceptive period before attempting the subsequent pregnancy, what matters is the resumption of the blood flow and that could vary depending on the individual case.

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