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Case Report

First case of twin pregnancy after vaginal radical trachelectomy in a Japanese woman

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ABSTRACT

Twin pregnancy after radical trachelectomy (RT) for uterine cervical cancer involves an extremely high risk of preterm premature rupture of the membrane (pPROM) and the following occurrence of preterm birth. Herein we report the first case of twin pregnancy after vaginal RT in a Japanese woman. The patient was a 36-year-old nulliparous woman. She became pregnant with twins one year after vaginal RT with laparoscopic lymphadenectomy and the following treatment with the use of in vitro fertilization/embryo transfer. Her pregnancy course was favorable with vaginal disinfection, bed rest, and administration of progesterone, ritodrine, and an ulinastatin vaginal suppository. At 31 weeks + 4 days of pregnancy, a scheduled cesarean section was performed. One baby weighed 1000g, with Apgar scores of 7/8, and the other 1100g, with Apgar scores of 8/9. Her postpartum course was also favorable. The mother remains without evidence of recurrence of the cancer at the time of this report.

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Introduction

Radical trachelectomy (RT) with pelvic lymphadenectomy, originally reported by Dargent et al., has now become a new treatment option for young patients in Japan with early invasive uterine cervical cancer who desire preservation of their fertility [1-3]. Recent data also indicate that the oncologic results following this operation are comparable to the results following standard radical hysterectomy (RH) [4]. We have performed this treatment modality for more than 70 patients and have experienced 20 pregnancies since 2003. Obstetrical management after RT is, however, very difficult for obstetricians. Uterine cervical conditions after RT are totally different from those after conization or normal pregnancy. The lack of mechanical support for the residual cervix, ascending infection and chorioamnionitis caused by disruption of the endocervical glands and reduced secretion of mucus can cause preterm premature rupture of the membrane (pPROM) and following

preterm birth. Therefore, for pregnancy after RT there are extremely high risks of early preterm abortion and preterm birth [5]. Under such conditions after RT, management of twin pregnancy can be a great challenge for obstetricians. There have only been 6 reports on 7 such patients published so far [6-11]. Here we report the successful outcome of a twin pregnancy at 31 weeks+4 days of pregnancy after vaginal RT. This is the first case of twin pregnancy after vaginal RT in Japan.

Case report

A 36-year-old Japanese woman, gravida 0, para 0, was diagnosed as having stage 1B1 uterine cervical cancer after examination of an abnormal Pap smear. As treatment options, radical hysterectomy and radical trachelectomy were presented to her and her husband. As they strongly desired to retain her fertility, vaginal RT with laparoscopic pelvic lymphadenectomy was performed based on the method of Dargent et al. Briefly, a rim of vaginal mucosa was delineated

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circumferentially and excised so that the anterior and posterior mucosae could cover the cervix. Vesicovaginal space was defined laterally on each side. Then the descending branches of the uterine arteries and the cardinal ligaments were cut at the level of Type II hysterectomy. After this procedure, the uterine cervix was amputated at 10mm below the internal os of the uterus. A prophylactic nylon suture was placed around the residual cervix to support cervical strength, and Strumdorf sutures were placed to cover the surface of the cervix.

Twelve months later, she became pregnant with twins after in vitro fertilization and embryo transfer (IVF-ET). From the shape of embryonic sacs, she was diagnosed as having dichorionic diamniotic (DD) twins from a single fertilized egg. She was then referred to our obstetrical section for the management of pregnancy. At 8 weeks of pregnancy, the cervical length of the “neo-cervix” was determined to be 20mm by transvaginal sonography. She was followed-up according to the follow-up method we reported previously [5]. Weekly administration of progesterone and daily disinfection with an ulinastatin vaginal suppository were performed under hospitalization. Administration of ritodrine is still allowed in Japan and prophylactic administration of ritodrine is also widely accepted under careful check of complications. Therefore, prophylactic administration of ritodrine was also introduced for this patient, too. Cervical length, bacterial vaginosis (BV), cervical elastase and fibronectin in the cervical mucosa were also checked weekly. As a result, her pregnancy course was favorable. Cervical length was maintained at more than 1cm over the pregnancy (Figure 1). Vaginal infection was also well controlled. At 31 weeks + 4 days of pregnancy, a scheduled cesarean section was performed. Babies were born weighing 1000g, with Apgar scores of 7/8, and 1100g, with Apgar scores of 8/9. The babies have been followed-up at our neonatal intensive care unit (NICU), and they are growing well without severe troubles. The mother’s postpartum course was also favorable, and she was discharged 1 week after the cesarean section. The mother remains without evidence of recurrence at the time of this report.

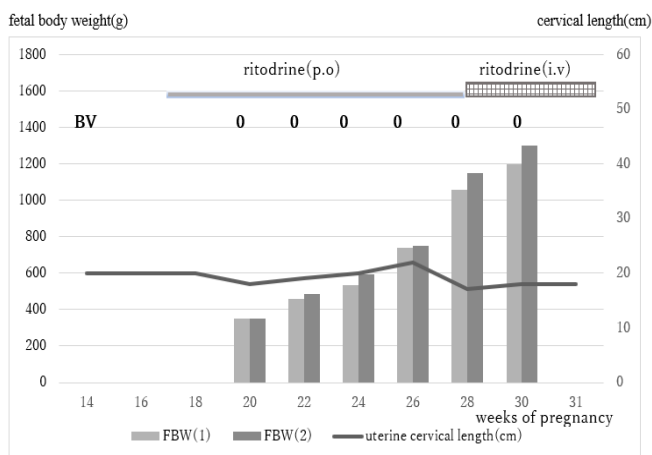


Figure 1: Pregnancy course of the patient.

Ritodrine (p.o) means oral administration of ritodrine. Ritodrine (i.v) means intravenous administration of ritodrine.

BV means the score of bacterial vaginosis. BV0 means no signs of bacterial vaginosis. FBW means fetal body weight measured with ultrasonography

Discussion

We reported the first successful outcome of a twin pregnancy (at 31 weeks 4 days) after vaginal RT in Japan. As (Table 1) demonstrates, there have been only 9 twin pregnancies after RT reported so far. Details of some of their pregnancy courses are unfortunately not available because they were written from the standpoint of oncological prognosis. As far as we know, most of them resulted in delivery in the late second trimester–early third trimester of pregnancy. Five of the eight patients underwent the emergency cesarean section before 31 weeks of pregnancy due to various maternal or fetal complications, and only three patients, including ours, could undergo scheduled cesarean section. In those emergency cases, the occurrence of preterm labor might have been the main cause of the termination. Thus, management of twin pregnancy after RT is very difficult for obstetricians.

There are no standard recommendations for the management of pregnancy after RT. Prevention of intrauterine infection that can lead to pPROM and the control of abdominal tension play important roles to achieve a better obstetrical prognosis [12].

As we reported previously, prophylactic cerclage placed at the time of RT seems to play important roles in preventing dilatation of the uterine cervix and the subsequent occurrence of pPROM [13]. We believe this prophylactic cerclage is inevitable, especially in cases of twin pregnancy. Five patients, including ours, underwent prophylactic cerclage at the time of RT, and three of them could undergo the scheduled cesarean section. In addition, prophylactic transabdominal cerclage (TAC) was also placed for patient 4. In cases of insufficient cerclage due to long duration after RT, or in cases where cerclage was not placed, TAC can be effective to support the function of the neo-cervix as we reported [14]. However, it is unclear whether another prophylactic additional cerclage improves the obstetrical prognosis of patients with twin pregnancy after RT. In addition, TAC during pregnancy has the risk of abortion or massive bleeding during the operation.

As it is well known, twin pregnancy itself is a high-risk pregnancy. It has a higher risk of preeclampsia. In cases of monochorionic diamniotic (MD) twins, it sometimes leads to twin-twin transfusion syndrome (TTTS). It also has risks of preterm labor and massive bleeding after delivery due to the large intrauterine volume. In such situations, there is no consensus among obstetricians as to how to decide the timing of termination. As we described in (Table 1), most cases resulted in emergent cesarean section. Considering the situation of twin pregnancies after RT, we set the timing of termination at 31 weeks of pregnancy. It goes without saying that it is preferable to extend the duration of pregnancy as long as possible. Our schedule of cesarean section at 31 weeks of pregnancy might have been too early to perform the operation. Babies who are born at this time of pregnancy have immature lung function and they usually have a risk of retinopathy. As their body weights are 1500-2000g, they usually need management in an NICU. However, as emergent cesarean section of twin pregnancy is a risk both for mother and babies, we set the timing of termination as the primary goal of this patient to avoid emergent situations after several conferences with neonatologists.

Management of twin pregnancy after RT is a rare condition. However, various assisted reproductive technologies (ARTs) are usually needed to conceive. Considering the higher possibility of multiple pregnancy with ART, we need to establish a standard follow-up modality for these patients [15].

Table 1: Characteristic of twin pregnancies after radical trachelectomy

case	age	stage	RT	cerclage	Reproductive Techniques	duration	membr.	treatment	delivery	cause of termination	of recurrence	Origin
1	39	1B1 SCC	VRT	+	IVF-ET	1 yrs	DD	Progesterone ritodrine	30+5	scheduled	—	This study
2	35	1B1 SCC	VRT	+	IUI	1 yrs	DD	bed rest progesterone	29+6	bag(+),pain	—	Yoon et al. ¹⁰
3	39	1B1 SCC	VRT	+	—	3 yrs	MD	bed rest	34+3	scheduled	—	Agarwal et al. ¹¹
4	33	1A1 adeno	VRT	+	IVF-ET	1.5 yrs	DD	TAC magnesium sulfate	30+5	bleeding	—	Lee et al. ⁸
5	31	1B1	VRT	n.a	n.a	n.a	MD	n.a	24+4	HELLP+TTTS	n.a	Bernardini et al. ⁷
6	31	1B1	VRT	n.a	n.a	n.a	n.a	n.a	26+2	pPROM	n.a	Bernardini et al. ⁷
7	n.a	1B1	VRT	n.a	Ovu.	n.a	n.a	n.a	24	Pain	—	Burnett et al. ⁶
8	36	1A1 SCC	ART	+	IVF-ET	5 yrs	n.a	n.a	36+1	Scheduled	—	Olawaiye et al. ⁹

RT:operative method of radical trachelectomy. VRT:vaginal radical trachelectomy, ART:abdominal radical trachelectomy.

duration: duration between the onset of pregnancy and the trachelectomy. n.a:not available membr.:types of twin pregnancy. DD:dichorionic diamniotic twin, MD: monochorionic diamniotic twin, IVF-ET:in vitro fertilization and embryo transfer. IUI:intra uterine insemination. ovu: simple ovulation check by ultrasonography. HELLP:hemolytic anemia, elevated liver enzyme,low platelet count syndrome. scheduled:scheduled cesarean section

Disclosure

The authors have no conflicts of interest to declare.

REFERENCES

- Dargent D, Martin X, Sacchetoni A, Mathevet P (2000) Laparoscopic vaginal radical trachelectomy: A treatment to preserve the fertility of cervical carcinoma patients. *Cancer* 88: 1877-1882. [Crossref]
- Kasuga Y, Nishio H, Miyakoshi K, Sato S, Sugiyama J, et al. (2016) Pregnancy outcomes after abdominal radical trachelectomy for early-stage cervical cancer: A 13-year experience in a single tertiary-care center. *Int J Gynecol Cancer* 26: 163-168. [Crossref]
- Sato S, Aoki D, Kobayashi H, Saito T, Nishimura R, et al. (2011) Questionnaire survey of the current status of radical trachelectomy in Japan. *Int J Clin Oncol* 16: 141-144. [Crossref]
- Diaz JP, Sonoda Y, Leitao MM, Zivanovic O, Brown CL, et al. (2008) Oncologic outcome of fertility-sparing radical trachelectomy versus radical hysterectomy for stage 1B1 cervical carcinoma. *Gynecol Oncol* 111: 255-260. [Crossref]
- Ishioka S, Endo T, Hayashi T, Baba T, Umemura K, et al. (2007) Pregnancy-related complications after vaginal radical trachelectomy for early-stage invasive uterine cervical cancer. *Int.J.Clin Oncol* 12: 350-355. [Crossref]
- Burnett AF, Roman LD, O Meara AT, Morrow CP (2003) Radical vaginal trachelectomy and pelvic lymphadenectomy for preservation of fertility in early cervical carcinoma. *Gynecol Oncol* 88: 419-423. [Crossref]
- Bernardini M, Barrett J, Seaward G, Covens A (2003) Pregnancy outcomes in patients after radical trachelectomy. *Am J Obstet Gynecol* 189: 1378-1382. [Crossref]
- Lee KY, Jun HA, Roh JW, Song JE (2007) Successful twin pregnancy after vaginal radical trachelectomy using transabdominal cervicoisthmic cerclage. *Am J Obstet Gynecol* 197: 5-6. [Crossref]
- Olawaiye A, Del Carmen M, Tambouret R, Goodman A, Fuller A, et al. Abdominal radical trachelectomy: Success and pitfalls in a general gynecologic oncology practice. *Gynecol Oncol* 112: 506-510. [Crossref]
- Yoon G, Choi S, Roh C, Choi D, Kim J, et al. (2010) Successful twin pregnancy by assisted reproductive technology after laparoscopic-assisted radical vaginal trachelectomy. *J.Womens Med* 3: 23-25.
- Agarwal N, Rana R, Gupta M, Ugwumadu A (2014) Successful delivery after a radical trachelectomy. *J Obstet Gynaecol* 34: 87.
- Ishioka S, Endo T, Hayashi T, Baba T, Umemura K, et al. (2007) Pregnancy-related complications after vaginal radical trachelectomy for early-stage invasive uterine cervical cancer. *Int J Clin Oncol* 12: 350-355. [Crossref]
- Kim M, Ishioka S, Endo T, Baba T, Akashi Y, et al. (2014) Importance of uterine cervical cerclage to maintain a successful pregnancy for patients who undergo vaginal radical trachelectomy. *Int J Clin Oncol* 19: 906-911. [Crossref]
- Ishioka S, Kim M, Mizugaki Y, Kon S, Ioyama K, et al. (2018) Transabdominal cerclage (TAC) for patients with ultra-short cervix after uterine cervix surgery and its impact on pregnancy. *J Obstet Gynaecol Res* 44: 61-66. [Crossref]
- Pinborg A (2005) IVF/ICSI twin pregnancies:risks and prevention. *Hum Reprod Update* 11: 575-593.[Crossref]