# Abdominal radical trachelectomy (ART) performed during pregnancy: a case report

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#### Summary

*Purpose of investigation:* Cervical cancer is one of the most frequent malignant diseases diagnosed during pregnancy. Abdominal or vaginal radical trachelectomies are fertility-preserving alternatives to radical hysterectomy for young women with early-stage cervical cancer that can be performed during ongoing pregnancy. *Material and Methods:* The authors report a pregnancy complicated by cervical cancer treated by abdominal radical trachelectomy (ART) at 16-17 gestational weeks with preservation of the concurrent pregnancy. *Results:* The pregnancy evolved normally and delivery occurred at 38-39 gestational weeks by elective caesarean section. *Conclusions:* Radical trachelectomy could be offered as an option for pregnant patients with early invasive cervical cancer. It may help women to avoid the triple losses of a desired pregnancy, fertility, and motherhood.

Key words: Cervical cancer; Pregnancy; Trachelectomy.

#### Introduction

Abdominal radical trachelectomy (ART) technique, imagined and described for the first time by the Romanian gynaecologist E. Aburel in 1956 [1], has been almost forgotten for four decades and "rediscovered" by Smith and Ungar's team in the 1990s [2]. Together with vaginal radical trachelectomy and laparoscopic pelvic lymphadenectomy described in 1994 by Dargent *et al.* [3], it represents a real option for women with cervical cancer Stages IA2-IB2 (IIA?), who deserve to preserve their fertility. Both techniques can be performed during ongoing pregnancy, but the successful reports so far are limited.

### **Case Report**

A 26-year-old primigravida, nullipara white female, was referred to the present clinic with the diagnosis of cervical cancer Stage IB1, and a 16 gestational weeks pregnancy. Her last period was on August  $22^{nd}$ , 2014. She had a HSIL smear test at the beginning of pregnancy and following a punch biopsy; the result showed an in situ carcinoma, with suspicion of invasion. A second biopsy demonstrated an invasive squamous carcinoma. On clinical examination a bleeding exophytic cervical mass of  $3\times3$  cm and a 16 gestational weeks pregnant uterus was discovered; parametria and rectum seemed normal. Obstetrical ultrasound demonstrated a 16-17 weeks pregnancy, with a female fetus with normal biometry, placenta inserted on the posterior uterine wall and normal amniotic fluid. Transrectal ultrasound demonstrated a cervical tumour of  $32\times38\times21$  mm, with invasion of less than half of the cervix in depth and without invasion into the parametria, bladder or rectum. Abdominal IRM described the same cervical lesion and pregnant uterus with no extraperitoneal enlarged lymph nodes and normal peritoneal cavity. The clinical diagnosis was Stage IB1 cervical carcinoma. Routine blood test investigations were normal, except of anemia with a haemoglobin level of 9.4 g/dl.

After informed consent, pre-anaesthetic evaluation and preoperative preparations, on December 17th, 2014, under a combined anesthetic technique using general anaesthesia with oro-tracheal intubation and an epidural catheter - for both intraoperative anesthesia and postoperative analgesia, the authors performed an ART with pelvic lymphadenectomy, with ligation of the left uterine artery and preservation of the right one, which was thicker, but without any intraoperative complications. A cerclage was performed before the completion of the anastomosis between the uterine isthmus and vagina. Intraoperatively, the frozen section of the upper part of the removed cervix showed no tumour at that level. The whole procedure lasted 295 minutes and the estimated blood loss was 500 ml. The postoperative recovery was complicated by a symptomatic right iliac lymphocele drained on ultrasound guidance on 12th postoperative day and an abdominal wound infection which required re-suturing on 18th day. The patient was discharged on the 22<sup>nd</sup> postoperative day. The final pathology report: cervix of 25×20×50 mm with vagina of 30 mm; grade 3 squamous carcinoma, invading eight of 24 mm of the cervix wall; no metastases into the parametria (right 55×24×24 mm, left 56×34×18 mm) and in the 23 right and 20 left removed lymph nodes.

The pregnancy evolved normally with monthly clinical, abdominal, and transvaginal ultrasound exams and Pap test sampled every three months. At 28 gestational weeks the patient was admitted in the hospital for a suspicion of premature labour, which was later confirmed. The pregnancy follow-up was even more careful during the last two months, with a clinical examination, ultrasound, and Doppler velocimetry performed every two weeks.

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Publication	Stage	Histology	GA at surgery (weeks)	Uterine arteries	Delivery	Outcome
Ungar et al., 2006 [9]	IB1	Squamous cell	7	Preserved	AB	NED
Ungar et al., 2006 [9]	IB1	Squamous cell	8	Preserved	AB	NED
Ungar et al., 2006 [9]	IB1	Squamous cell	9	Preserved	38	NED
Ungar et al., 2006 [9]	IB1	Squamous cell	13	Preserved	AB	NED
Ungar et al., 2006 [9]	IA2	Squamous cell	18	Preserved	39	NED
Mandic et al., 2009 [11]	IB1	Squamous cell	19	NA	36	NED
Abu-Rustum et al., 2009 [10]	IB1	Lymphoepithelial	15	Left ligated	39	NA
Enomoto et al., 2011 [13]	IB1	Squamous cell	15	Right ligated	37	NED
Karateke et al., 2010 [12]	IB1	Squamous cell	22	Bilateral ligated	AB	NA
Aoki et al., 2014 [14]	IB1	Squamous cell	17	Preserved	38	NED
Capîlna (this case report)	IB1	Squamous cell	17	Left ligated	38	NED

Table 1. — Reported abdominal radical trachelectomy patients: stage, gestational age at surgery, obstetrical, and oncology outcome.

GA: gestational age; AB: abortion; NA: not applicable; NED: no evidence of disease;

The patient delivered at 38-39 gestational weeks by elective caesarean section on May 17<sup>th</sup>, 2015 a male weighing 2,950 grams and with an Apgar score of 9/1 minute. The procedure was complicated by a very intense hemorrhage arising from the presacral area, after uterine mobilization. She received blood and plasma transfusion, intra- and postoperatively. The recovery was uneventful for both mother and newborn, with hospital discharge on the sixth postoperative day. At this moment, two months after delivery, the mother and baby are both fine, with normal follow-up.

#### Discussion

The authors conducted a literature review of articles on this subject through a Medline search for articles published in English or French. The main search terms were *cervical cancer*, *pregnancy*, and *trachelectomy*. At this moment, 21 cervical cancer patients, including the present (four Stage IA2, 16 IB1, and one IB2) who underwent radical trachelectomy during pregnancy have been reported. Of these, ten were performed by vaginal route [4-8] and 11 were abdominal radical trachelectomies (Table 1) [9-14].

Regarding ART, the surgery was performed between seven and 22 gestational weeks in all 11 patients. Four pregnancy ended in spontaneous abortion in the first 16 days after the procedure. Three of the four patients who underwent ART earlier than 14 weeks of gestation had miscarriages. This suggests that fetuses in the first trimester may not be able to tolerate the ART procedures [14]. The ART treatment at 22 weeks of gestation in another patient resulted in intrauterine fetal death four hours after the surgery, but both uterine arteries were ligated during trachelectomy and the surgeons performed an extensive surgery, including a para-aortic lymphadenectomy. Although the fetal autopsy revealed no abnormal findings, the placenta showed hypoxic changes without any sign of abruption. This case suggests that blood supply delivered only by the ovarian arteries might be insufficient in late gestation, resulting in loss of the pregnancy. Also, an extensive surgery with excessive uterine manipulation could be deleterious for the ongoing pregnancy.

In terms of oncologic result, all 11 patients who underwent a ART had squamous cell carcinoma (ten cases) and a poorly differentiated lymphoepithelioma-like cervical cancer for one. All patients, except one staged IA2, were in Stage IB1. No patient had positive lymph nodes at the time of ART. All patients were free of disease during follow-up at the time of this report. Aside from the technically challenging procedure, ART during pregnancy was associated with significant blood loss (200-2,510 ml) and prolonged surgery (3.5–7.5 hours). Nonetheless, this type of surgery is addressed in young patients, and, even more, the pregnancy induced hypervolemia allows to better compensate a more significant intraoperative blood loss. The present authors do not consider that the risk of bleeding is a reason to avoid the surgery. Also, as it is known from radical abdominal hysterectomy performed during early pregnancy or at the time of cesarean section for an invasive cervical cancer, the pregnancy induced changes in the pelvis allow an easy dissection and mobilization of pelvic anatomical structures.

The rate of fetal loss after this procedure was high (four fetal losses occurred 0–16 days after surgery). It appears safer and with better obstetrical results to postpone the ART to the second trimester of pregnancy. All the six patients, including the present, who underwent surgery between 15 and 19 weeks of gestation, gave birth to healthy live babies. To ensure a better blood supply to the pregnant uterus, both uterine arteries, or at least one, must be preserved, if possible, when performing the ART during pregnancy.

## Conclusion

The present authors believe that it is important to offer this surgical option to pregnant patients with invasive cervical cancer. Best pregnancy period to perform a radical trachelectomy looks to be the early second trimester. Detailed investigation of similar cases would reveal the efficacy and safety of this procedure, although it is a technically challenging approach and the importance of experience and centralization should be emphasized. It may be possible that radical trachelectomy during pregnancy can help women to avoid the triple losses of a desired pregnancy, fertility, and motherhood. On the contrary, new studies regarding the safety of chemotherapy during pregnancy are needed.

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