



Case report

Abdominal radical trachelectomy for vaginal cancer – A case report



Mari Matsuoka*, Rumiko Yamamoto, Natsuki Tsuji, Koichi Terakawa, Tadayoshi Nagano

Department of Obstetrics and Gynecology, Kitano Hospital, The Tazuke Kofukai Medical Research Institute, 2-4-20 Ohgimachi, Kita-ku, Osaka 530-8480, Japan

1. Introduction

Primary vaginal cancer accounts for approximately 3% of all gynecological malignancies. It is very common in elderly women, while very rare in the young. The present report describes the treatment methods and clinical experience in a young woman with primary vaginal cancer in whom fertility was preserved.

2. Case report

The patient was a married 34-year-old para 1 Japanese woman. Pap smear for cervical screening revealed HSIL (class IIIb). Thus, she was referred to our hospital in January of year X. Histologic examination of the biopsy revealed mild cervical dysplasia. Therefore, she was monitored regularly. She moved overseas, and a routine test performed there showed a vaginal tumor. Consequently, she was examined again at our hospital during July of year X + 1. Speculum examination showed a papillary tumor in one third of the vaginal cranial region, extending from the posterior wall to the left lateral wall. Colposcopy showed no spread of the tumor to the cervical region and no cervical tumors (Fig. 1). Rectal examination showed no parametrial invasion. Vaginal wall cytology revealed VAIN 3 (class IIIb) and vaginal wall biopsy revealed non-keratinizing squamous cell carcinoma. Cervical cytology revealed HSIL (class IIIb), and cervical biopsy revealed CIN3 (severe dysplasia). Examination of the endometrium showed no abnormalities. The results for tumor markers were as follows: CEA: 0.5 ng/ml, CA 19-9: 5.8 U/ml, CA 125: 11 U/ml, and SCC: 1.1 ng/ml. Although the vaginal tumor was not detected by transvaginal ultrasonography, pelvic MRI showed a 4.6 × 0.9 cm tumor, extending from the vaginal posterior wall to the left lateral wall, with a moderate signal on T2-weighted images and an intense signal on diffusion-weighted images. There were no signs of paravaginal invasion (Fig. 2). PET-CT showed increased accumulation of FDG in the vaginal tumor with SUV-max of 10.92 in the early phase and 11.24 in the late phase. There were no signs of lymph node or distant metastases. Based on the above findings, a diagnosis of stage I vaginal squamous cell carcinoma (FIGO Committee on Gynecologic Oncology, 2009) was made. Although she was parous, she and her family expressed a strong desire for fertility preservation. We obtained informed consent, and then administered two cycles of intra-arterial chemotherapy, each consisting of 100 mg of

cisplatin (CDDP) and 10 mg of mitomycin C (MMC), followed by abdominal radical trachelectomy (ART), vaginal wall resection, and pelvic lymph node dissection. It was also decided that fertility preservation would be abandoned, if the chemotherapy showed insufficient efficacy.

Angiography showed a larger right uterine artery. There was a deeply stained region, which was considered to be the vaginal tumor, in the lower left part of the uterus. In addition, there was a deeply stained region in a segment of the right uterine artery, which was thought to be a tumor. Based on these findings, 100 mg of CDDP and 10 mg of MMC were administered via the uterine artery in a left/right ratio of 6:4.

One month after intra-arterial chemotherapy, colposcopy showed a visible decrease in tumor size, and MRI showed a 42% decrease in size (from 4.6 × 0.9 cm to 3.4 × 0.7 cm). A judgment of partial response was made, and we decided to administer the second chemotherapy cycle. After the second cycle, MRI showed a further decrease in tumor size (by 56% compared to the pre-treatment size (from 4.6 × 0.9 cm to 3.0 × 0.6 cm)). Surgery was performed one month after the second chemotherapy cycle, using the same procedure as that used for ART to treat cervical cancer. Intra-operative frozen section of the vaginal wall resection stump was negative. In addition, the result of intraoperative ascites cytology was negative. The bilateral external iliac lymph nodes and left obturator lymph node were mildly enlarged, but were negative on frozen section. Therefore, ART was performed in combination with vaginal wall resection and pelvic lymph node dissection. Operative time was 6 h and 58 min, and blood loss was 1170 g. The vaginal tumor was excised from the posterior vaginal wall to the left lateral wall, and its size was 2 × 1 × 0.5 cm (Fig. 3). Loupe imaging also showed a tumor in the vaginal wall, but without spread to the cervical region, so it was concluded to be a primary vaginal tumor (Fig. 4). No signs of malignancy were found in the cervix, but signs of keratinizing squamous cell carcinoma were found in the vaginal wall over an area of 2 cm in the horizontal direction and 1.3 cm in the vertical direction with an invasion depth of 0.5 cm. No vascular invasion was found, and tests on the vaginal stump were negative. Based on these findings, we diagnosed this tumor as stage I vaginal squamous cell carcinoma (FIGO Committee on Gynecologic Oncology, 2009, yp T1N0M0).

No postoperative complications occurred and menstruation was normal. No adjuvant treatment was administered, and no recurrence was reported up until 18 months postoperatively.

* Corresponding author.

E-mail address: mari-matsuoka@kitano-hp.or.jp (M. Matsuoka).

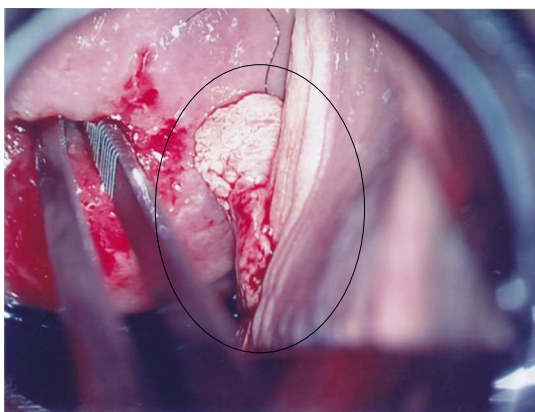


Fig. 1. Colposcopic findings at the initial examination: A papillary tumor is seen occupying one third of the vaginal cranial region extending from the posterior wall to the left lateral wall (black ellipse). However, it does not extend to the cervical region, and no cervical tumor was found. The forceps is inserted through the external uterine orifice.

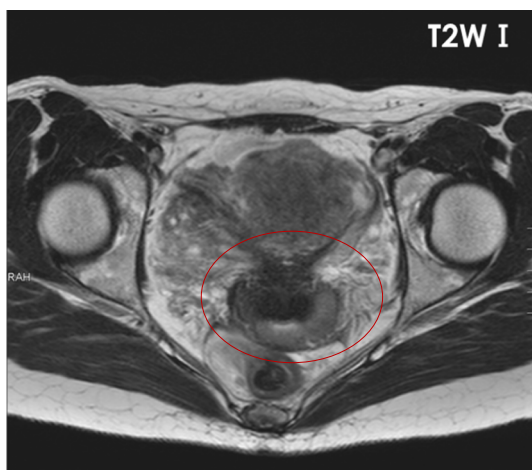


Fig. 2. MRI: A tumor with a moderate signal on T2-weighted imaging, extending from the vaginal posterior wall to the left lateral wall (red ellipse), is seen. However, it does not extend to the cervical region and there are no signs of paravaginal invasion. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

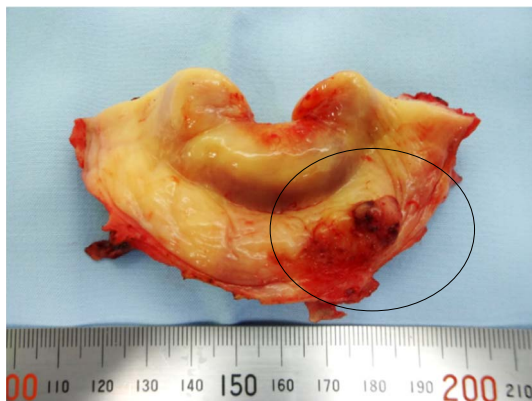


Fig. 3. Excised samples: A papillary tumor, measuring 2 × 1 × 0.5 cm, is found to extend from the vaginal posterior wall to the left lateral wall.

3. Discussion

Vaginal cancer was first reported by [Graham et al. \(1952\)](#), but the number of reports on this rare tumor, which accounts for only 3% of all gynecological malignancies ([Siegel et al., 2015](#)), has been small. The

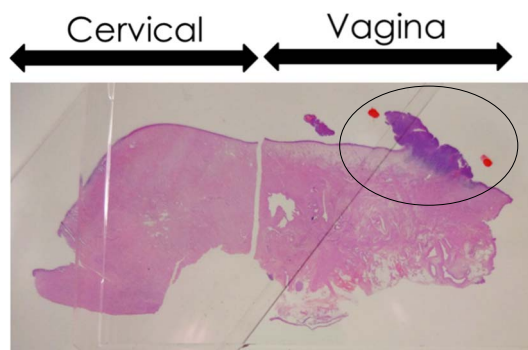


Fig. 4. Loupe image: A tumor is found in the vaginal wall, but does not extend to the cervical region.

most common type of this tumor is squamous cell carcinoma ([Cerasman et al., 1998](#)). The mean age at diagnosis is 65, and only 5% of cases are diagnosed before the age of 40 ([Shah et al., 2009](#)). Surgery and radiotherapy are recommended for treating stage I vaginal cancer and the 5-year survival rate tends to be higher with surgery (77%) compared to radiotherapy alone (68%) ([Tjalma et al., 2001](#)). The probability of lymph node metastasis with stage I vaginal cancer is 6–16% ([Davis et al., 1991](#); [Pride et al., 1979](#); [Stock et al., 1995](#)) and the risk of metastasis increases with increasing tumor diameter, increasing invasion depth, and decreasing histological differentiation ([Perez et al., 1999](#)).

In the present case, the tumor was restricted to the vaginal wall, but was relatively large, measuring 4.6 × 0.9 cm. There has been a recent increase in the number of reports on the use of radical trachelectomy with neoadjuvant chemotherapy to treat cervical cancer involving large tumors of a diameter of 2 cm or more, but the safety of this approach has not been established ([Lanowska et al., 2014](#); [Pareja et al., 2015](#)). In addition, [Chirag et al. \(2009\)](#) reported that the prognosis is significantly poorer with tumors over 4 cm in diameter.

On the other hand, [Benedetti et al. \(2008\)](#) reported favorable results in 11 patients with preoperative tumor diameters between 4.1 cm and 7.2 cm who were treated with neoadjuvant chemotherapy and radical hysterectomy for stage II vaginal cancer. Their survival rate (mean follow-up period, 75 months; range, 12–103 months) was 91% and the disease-free survival rate was 73%. The number of patients included in Benedetti's study was small, but the findings suggest the possible effectiveness of neoadjuvant chemotherapy for treating advanced vaginal cancer.

Although our patient's pre-treatment tumor diameter was over 4 cm, the tumor was restricted to the vaginal wall and there were no signs suggesting lymph node or distant metastases. Therefore, we concluded that if the tumor responded to neoadjuvant chemotherapy, it would be possible to perform complete surgery, maintaining a negative status at the resection stump. During surgery, an incision was first made into the vaginal anterior wall, and tumor removal was visually confirmed. Grossly, the tumor free margin measured approximately 3 mm only, so additional vaginal resection was performed, and frozen section confirmed the negative status of the vaginal stump. The negative status of the vaginal stump was also confirmed with permanent samples.

Additionally, as neither lymph node metastasis, vascular invasion nor parametrial invasion were found, no adjuvant treatment was administered, and no signs of recurrence were detected up until 18 months postoperatively by cervical and vaginal smears, systemic CT and pelvic MRI.

Our patient developed asymptomatic vaginal cancer that was detected incidentally during follow-up for cervical dysplasia. The proportion of asymptomatic patients with vaginal cancer is 5–10%, and these are detected during screening for cervical cancer or on pelvic examination ([Di Donato et al., 2012](#)). Furthermore, HPV infection has been suggested as a risk factor for vaginal cancer. According to [Sinno](#)

et al. (2014), the HPV-positivity rate in the patients aged 60 or younger with vaginal cancer was high at 91%, and the virus was HPV-16 or – 18 in 77.3% of cases. No HPV test was performed in our patient, but it is possible that she was positive for high-risk HPV.

The occurrence of cervical cancer is increasing in younger women, and this is becoming a societal problem. As patients with vaginal cancer are also often HPV-positive, there are concerns about the potential for an increase in vaginal cancer in younger women. There is a worldwide recommendation for HPV vaccination to protect against cervical cancer, and it can be expected that this will also serve to protect against vaginal cancer.

When young people develop malignancies of the reproductive organs, in addition to the prognosis, it is also important to preserve fertility. Our patient was treated with neoadjuvant chemotherapy, ART, vaginal wall resection, and pelvic lymph node dissection and her fertility was preserved. To the best of our knowledge, this report is the first case of abdominal radical trachelectomy for vaginal cancer. The prognosis with such treatment is also unknown, so careful monitoring will be needed.

Conflict of interest statement

All authors declare that there are no conflicts of interest.

References

- Benedetti, P.P., et al., 2008. Neoadjuvant chemotherapy followed by radical surgery in patients effected by vaginal carcinoma. *Gynecol. Oncol.* 111 (2), 307–311.
- Cerasman, W.T., et al., 1998. The National Cancer Data Base report on cancer of the vagina. *Cancer* 83 (5), 1033–1040.
- Chirag, A., et al., 2009. Factors affecting risk of mortality in women with vaginal cancer. *Obstet. Gynecol.* 113 (5), 1038–1045.
- Davis, K.P., et al., 1991. Invasive vaginal carcinoma: analysis of early stage disease. *Gynecol. Oncol.* 42 (2), 131–136.
- Di Donato, V., et al., 2012. Vaginal cancer. *Crit. Rev. Oncol. Hematol.* 81 (3), 286–295.
- FIGO Committee on Gynecologic Oncology, 2009. Current FIGO staging for cancer of the vagina, fallopian tube, ovary, and gestational trophoblastic neoplasia. *Int. J. Gynecol. Obstet.* 105, 3–4.
- Graham, J.B., et al., 1952. Earlier detection of recurrent cancer of the uterine cervix by vaginal smear. *Am. J. Obstet. Gynecol.* 64 (4), 908–914.
- Lanowska, M., et al., 2014. Radical vaginal trachelectomy after laparoscopic staging and neoadjuvant chemotherapy in women with early-stage cervical cancer over 2 cm: oncologic, fertility, and neonatal outcome in a series of 20 patients. *Int. J. Gynecol. Cancer* 24 (3), 586–593.
- Pareja, R., et al., 2015. Immediate radical trachelectomy versus neoadjuvant chemotherapy followed by conservative surgery for patients with stage IB1 cervical cancer with tumors 2cm or larger: a literature review and analysis of oncological and obstetrical outcomes. *Gynecol. Oncol.* 137 (3), 574–580.
- Perez, C.A., et al., 1999. Factors affecting long-term outcome of irradiation in carcinoma of the vagina. *Int. J. Radiat. Oncol. Biol. Phys.* 44 (1), 37–45.
- Pride, G.L., et al., 1979. Primary invasive squamous carcinoma of the vagina. *Obstet. Gynecol.* 53 (2), 218–225.
- Shah, C.A., et al., 2009. Factors affecting risk of mortality in women with vaginal cancer. *Obstet. Gynecol.* 113 (5), 1038–1045.
- Siegel, R.L., et al., 2015. Cancer statistics. *CA Cancer J. Clin.* 65 (1), 5.
- Sinno, A.K., et al., 2014. Human papillomavirus genotype prevalence in invasive vaginal cancer from a registry-based population. *Obstet. Gynecol.* 123 (4), 817–821.
- Stock, R.G., et al., 1995. A 30-year experience in the management of primary carcinoma of the vagina: analysis of prognostic factors and treatment modalities. *Gynecol. Oncol.* 56 (1), 45–52.
- Tjalma, W.A., et al., 2001. The role of surgery in invasive squamous carcinoma of the vagina. *Gynecol. Oncol.* 81 (3), 360–365.