

Prolonged survival after episiotomy recurrence of cervical cancer complicating pregnancy

I. Hafeez¹, B.D. Lawenda¹, J.M. Schilder², P.A.S. Johnstone¹

¹Department of Radiation Oncology, ²Division of Gynecologic Oncology, Indiana University School of Medicine, Indianapolis, IN (USA)

Summary

Background: We report a case of recurrent cervical cancer in an episiotomy scar and the late treatment-related sequelae. **Case:** Cervical cancer was diagnosed following a vaginal delivery, and was treated with surgery and radiotherapy. The patient developed a recurrence in her episiotomy scar, and was treated with chemoradiation. She remains without evidence of disease ten years later. **Conclusion:** Successful treatment of recurrent cervical cancer with chemoradiation is possible, but may be associated with significant normal tissue toxicity.

Key words: Episiotomy; Cervical cancer; Pregnancy complication; Surgery; Radiation therapy.

Introduction

Although the exact incidence and prevalence of cancer during pregnancy remains unknown, reliable estimates suggest that it is uncommon, occurring at a frequency of between one to two new cases in every 1,000 pregnancies [1]. More specifically, the incidence of cervical carcinoma during pregnancy is estimated to be 1.2 cases per 10,000 pregnancies [2]. However there have been only 17 cases of cervical cancer recurrence at the site of an episiotomy in the English language literature (Table 1). The majority of cases (14 of 17) recurred within five months of delivery; many were rapidly fatal. We discuss the long-term course of treatment for – and sequelae of – a woman with cervical adenosquamous carcinoma diagnosed at the time of delivery with subsequent recurrence at the episiotomy scar. The case highlights an exceptionally unique example of long-term survival following an episiotomy recurrence of cervical cancer that was treated with chemoradiation alone and without excision and the issues surrounding quality of life that such survival entails.

Case Report

A 35-year-old female, G3P3A3, received a diagnosis of squamous cell carcinoma (IBG2) of the cervix at the time of delivery at another medical treatment facility. Following an otherwise normal vaginal delivery, the patient underwent radical hysterectomy and postoperative cobalt radiotherapy. She received 50 Gy to the whole pelvis using opposed anteroposterior fields, with the placement of a midline block after 38 Gy. More detailed records of her initial treatment were not available. Five months following treatment, the patient was found to have a perineal recurrence of her cancer at the site of the episiotomy. An exploratory laparotomy, with pelvic lymph node dissection, at the time was negative. She received 45 Gy using small anteroposterior photon fields inferior to the area demarcated by her prior treatment tattoos and existing subcutaneous

fibrosis. A 16.2 Gy electron boost was then delivered using the en face technique to perineal residual disease at that point, to a total dose of 61.2 Gy with five cycles of concurrent 5-fluorouracil and mitomycin. Within three months of completion of chemoradiation, the patient developed dyspareunia, proctitis, and a persistent, tender ulcer of the perineal body. Excision was negative for malignancy; however, the patient developed necrosis of the perineal body with persistent tenesmus and rectal bleeding. Over the ensuing years, she experienced dyspareunia and post-coital bleeding, and subsequent rectovaginal fibrosis. The patient declined neovagina formation, and these complications ultimately were managed with a sigmoid colectomy and diverting ileostomy. At ten years following hysterectomy, the patient developed recurrent episodes of perineal cellulitis, and an obstructive uropathy considered secondary to bladder dysfunction from radiation-induced changes with bilateral hydronephrosis, hydroureters, and marked postvoid residual volumes. These urologic toxicities require continued management. She remains without evidence of disease recurrence.

Discussion

Cervical cancer complicating pregnancy is a rare event. Even more infrequent is cancer recurrence at the site of an episiotomy scar. Seventeen such cases have been reported to date, with patient ages ranging from 21 to 37 (Table 1). Survival times vary, but seem to improve notably with aggressive therapy of the recurrent lesion typically consisting of excision with chemoradiation or brachytherapy in combination with external beam radiation. We found only four reported cases where patients with episiotomy recurrences were treated using chemoradiation alone [7, 9, 11, 13]. In three of these, the patients died of disease within three years of recurrence. Our patient thus is an anomaly: she was treated using salvage chemoradiation alone without excision, and has survived more than 120 months after her recurrence.

To avoid a theoretical risk of dystocia and massive bleeding from the tumor mass, cesarean section as a route of delivery has been a prevailing recommendation for

Revised manuscript accepted for publication June 22, 2010

Table 1. — Cases of episiotomy recurrences of cervical cancer from the literature.

References	Patient age	Histology	Stage of cervical cancer	Time post-delivery of episiotomy implant cancer	Treatment of episiotomy implant	Survival following treatment of episiotomy implant
Burgess [3]	33	S	IB	28 months	abdominoperineal resection	> 6 months
Copeland <i>et al.</i> [4]	32	A	IB	6 months	excision and radiotherapy	> 5 years
Copeland <i>et al.</i> [4]	32	A	IB	5 months	excision and radiotherapy	10 months
Gordon <i>et al.</i> [5]	24	S	IB	2.5 month	excision and radiotherapy	> 3.5 years
Van Dam <i>et al.</i> [6]	29	S	IIA	2.25 months	chemotherapy and radiotherapy	not available
Van Dam <i>et al.</i> [6]	30	A	IB	5 months	excision and radiotherapy	> 10 years
Khalil <i>et al.</i> [7]	32	S	IIIB	3 months	chemotherapy and radiotherapy	1 year
Cliby <i>et al.</i> [8]	37	S	IB	1.75 months	combination chemotherapy	6 months
Cliby <i>et al.</i> [8]	31	S	IB	2 years	chemotherapy, excision, and radiotherapy	1 year
Cliby <i>et al.</i> [8]	21	S	IB	4.25 months	radiotherapy	6 months
Cliby <i>et al.</i> [8]	34	S	IB	4 months	excision and radiotherapy	1 year
Van den Broek <i>et al.</i> [9]	29	S	IA1	1.75 months	chemotherapy and radiotherapy	3 years
Goldman [10]	35	A	IB	5.5 years	radiotherapy	6 months
Amanie <i>et al.</i> [11]	30	A	IIIA	1.5 years	radiotherapy and chemotherapy	1 year
Heron <i>et al.</i> [12]	32	A	IB1	8.7 years	radiotherapy	> 10 months
Baloglu <i>et al.</i> [13]	36	S	IIIA	8 months	radiotherapy and chemotherapy	>1 year
Neumann <i>et al.</i> [14]	35	AS	IB2	5 weeks	radiotherapy and chemotherapy	8 months
Hafeez <i>et al.</i>	35	S	IB	10 months	radiotherapy and chemotherapy	> 10 years

A = Adenocarcinoma, S = Squamous Carcinoma, AS = Adenosquamous Carcinoma.

pregnancies complicated by cervical cancer since the 1960s [15]. However proof of such benefit is difficult to determine from the literature. It is not known whether delivery by cesarean section carries a risk for tumor implantation in the abdominal scar or for metastases to regional lymphatics, and at least one case suggests the possibility of cervical cancer recurrence in the scar of a previous cesarean section [16]. Furthermore, the prognosis for cervical cancer diagnosed during pregnancy remains in dispute, with Hopkins *et al.* reporting no decrement in survival of patients with Stage IB cervical cancer associated with pregnancy [17]. A case-control study in women diagnosed postpartum done in 2000, however, showed an increased risk of recurrent disease, with generally poorer outcome, especially after vaginal delivery, versus women diagnosed during pregnancy [18].

Chronic radiation effects to tissue at risk of scarring and thus poor vascularity may be significant. We have previously discussed this effect in treating skin cancers of the lower extremity in the elderly [19]. The result was similarly dramatic here in a perineal body biopsied several months after chemoradiation: necrosis and persistent rectovaginal communication distally requiring colostomy.

In conclusion, recurrence of cervical cancer at the site of an episiotomy scar is rare. Long-term prognosis varies greatly, with increased survival potentially favored by aggressive therapy, including chemoradiation. We report a case of long-term survival greater than ten years following chemoradiation. This case illustrates the importance of inspecting the perineal area of a patient diagnosed with cervical cancer during pregnancy, especially after vaginal delivery and when an episiotomy has been performed. In the event of cervical cancer diagnosed during pregnancy with unavoidable vaginal delivery and episiotomy, regular reexamination of the scar is prudent.

References

- [1] Wingo P.A., Tong T, Bolden S.: "Cancer statistics". *CA Cancer J. Clin.*, 1995, 45, 8.
- [2] Duggan B., Mudderspach L.I., Roman L.D., Curtin J.P., d'Alaing G. 3rd, Morrow C.P.: "Cervical cancer in pregnancy: reporting on planned delay in therapy". *Obstet. Gynecol.*, 1993, 82, 598.
- [3] Burgess S.P., Waymont B.: "Implantation of a cervical carcinoma in an episiotomy site. Case report". *Br. J. Obstet. Gynaecol.*, 1987, 94, 598.
- [4] Copeland L.J., Saul P.B., Sneige N.: "Cervical adenocarcinoma: tumor implantation in the episiotomy sites of two patients". *Gynecol. Oncol.*, 1987, 28, 230.
- [5] Gordon A.N., Jensen R., Jones H.W. 3rd: "Squamous carcinoma of the cervix complicating pregnancy: recurrence in episiotomy after vaginal delivery". *Obstet. Gynecol.*, 1989, 73, 850.
- [6] Van Dam P.A., Irvine L., Lowe D.G., Fisher C., Barton D.P., Shepherd J.H.: "Carcinoma in episiotomy scars". *Gynecol. Oncol.*, 1992, 44, 96.
- [7] Khalil A.M., Khatib R.A., Mufarrij A.A., Tawil A.N., Issa P.Y.: "Squamous cell carcinoma of the cervix implanting in the episiotomy site". *Gynecol. Oncol.*, 1993, 51, 408.
- [8] Cliby W.A., Dodson M.K., Podratz K.C.: "Cervical cancer complicated by pregnancy: episiotomy site recurrences following vaginal delivery". *Obstet. Gynecol.*, 1994, 84, 179.
- [9] Van den Broek N.R., Lopez A.D., Ansink A., Monaghan J.M.: "'Microinvasive' adenocarcinoma of the cervix implanting in an episiotomy scar". *Gynecol. Oncol.*, 1995, 59, 297.
- [10] Goldman N.A., Goldberg G.I.: "Late recurrence of squamous cell cervical cancer in an episiotomy site after vaginal delivery". *Obstet. Gynecol.*, 1003, 101, 1127.
- [11] Amanie J., Pearcey R.G., Honore L., Sloboda R.: "Metastatic adenocarcinoma of the cervix in a delivery-induced traumatic lower vaginal tear". *Gynecol. Oncol.*, 2005, 96, 857.
- [12] Heron D.E., Axtel A., Gerszten K.: "Villoglandular adenocarcinoma of the cervix recurrent in an episiotomy scar: a case report in a 32-year-old female". *Int. J. Gynecol. Cancer*, 2005, 15, 366.
- [13] Baloglu A., Uysal D., Aslan N., Yigit S.: "Advanced stage of cervical carcinoma undiagnosed during antenatal period in term pregnancy and concomitant metastasis on episiotomy scar during delivery: a case report and review of the literature". *Int. J. Gynecol. Cancer*, 2007, 17, 1155.
- [14] Neumann G., Rasmussen K.L., Petersen L.K.: "Cervical adenosquamous carcinoma: tumor implantation in an episiotomy scar". *Obstet. Gynecol.*, 2007, 110, 467.

- [15] Williams T.J., Brack C.B.: "Carcinoma of the cervix in pregnancy". *Cancer*, 1964, 17, 1486.
- [16] Greenlee R.M., Chervenak F.A., Tovell H.M.: "Incisional recurrence of a cervical carcinoma. Report of a case". *JAMA*, 1981, 246, 69.
- [17] Hopkins M.P., Morley G.W.: "The prognosis and management of cervical cancer associated with pregnancy". *Obstet. Gynecol.*, 1992, 80, 9.
- [18] Sood A.K., Sorosky J.I.: "Cervical cancer diagnosed shortly after pregnancy: prognostic variable and delivery routes". *Obstet. Gynecol.*, 2000, 95, 832.
- [19] Dupree M.T., Kitley R.A., Weismantle K., Panos R., Johnstone P.A.S.: "Radiation therapy for treatment of Bowen's disease: lessons for lesions of the lower extremity". *J. Am. Acad. Dermatol.*, 2001, 45, 401.
- [20] Hacker N.F., Berek J.S., Lagasse L.D., Charles E.H.: "Carcinoma of the cervix associated with pregnancy". *Obstet. Gynecol.*, 1982, 28, 230.
- [21] Mikuta J.J.: "Invasive carcinoma of the cervix in pregnancy". *South Med. J.*, 1967, 60, 823.
- [22] Ra K.: "Factors affecting the prognosis of cancer of the cervix in pregnancy". *Am. J. Obstet. Gynecol.*, 1961, 82, 45.
- [23] Pradhan S., Asthana A.K., Sharan G.K.: "Recurrence of carcinoma cervix in the scar of previous cesarean section: a case report". *Int. J. Gynecol. Cancer*, 2006, 16, 900.

Address reprint requests to:
P.A.S. JOHNSTONE, M.D.
Department of Radiation Oncology
Indiana University School of Medicine
535 Barnhill Dr (RT041)
Indianapolis, IN 46202 (USA)
e-mail: pajohnst@iupui.edu