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Case Report

Pregnancy following vulvar squamous cell carcinoma: a report of two cases

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Pregnancy following squamous cell carcinoma of the vulvar is rare. Its rarity is reflected by a paucity of cases reported in the literature. We report two cases of pregnancy following diagnosis and treatment for vulvar squamous cell carcinoma, and review eleven prior reported cases. In successfully treated vulvar cancer subsequent pregnancy is not shown to increase the risk of disease recurrence, and there appears to be no deleterious effects during the antenatal period. It is possible, when considering prior reports, that prior vulvectomy may increase the likelihood of delivery by caesarean section, though modifications in the surgical management of vulvar carcinoma may have decreased this risk.

Key Words: Vulvar cancer, Squamous cell carcinoma, Radical vulvectomy, Pregnancy

INTRODUCTION

Reports of pregnancy following treatment for squamous cell carcinoma of the vulva are rare. In recent years an increased incidence of vulvar cancer in younger women has been observed,[1] with a concomitant increase in the incidence of human papillomavirus, vulvar intraepithelial neoplasia (VIN) and human immunodeficiency virus. Obstetricians and gynaecological oncologists are progressively more likely to encounter patients presenting in pregnancy following treatment for vulvar cancer. Given the sparse literature on this subject, we present two cases from our unit highlighting some of the difficulties that arise in patient management.

CASE REPORTS

1. Case 1

A 29 year old parous (G2P1) smoker presented in August 1989 with a twelve-month history of vulvar pruritis. She had a prior history of recurrent genital herpes, genital warts, and VIN 3. Examination revealed an extensive, thick, white lesion with coarse punctuation over the entire right labium minus. Vulvar biopsy confirmed VIN 3 with the presence of early invasion. A radical vulvectomy with bilateral groin node dissection was performed with subsequent vulvar skin grafting. Histology revealed carcinoma in situ on a background of VIN 3 (VIN 3 extending to the excision margin). Lymph nodes were negative and a diagnosis of FIGO stage I squamous cell carcinoma (SCC) of the vulva was made. Seven months later laser treatment and a (later reversed) loop ileostomy were performed for extensive perianal intraepithelial neoplasia.

Two years following primary diagnosis she presented at eleven weeks gestation. In view of extensive vulvar scarring delivery by caesarean section was planned. The antenatal period was routine and a live healthy infant was delivered by elective caesarean section at 38 weeks gestation. She has since undergone two further local excisions for recurrent anal intraepithelial neoplasia (AIN) and remains alive and well and free of recurrence twenty years post primary diagnosis.

2. Case 2

A 36-year old nulliparous, non-smoker presented in January 2000 with a history of vulvar pruritis. She had a prior history of genital herpes and genital warts. Examination revealed a large area of thickened pigmented skin affecting the labia minora, posterior vaginal fourchette, and perineum with extension towards the anal margin. Proctoscopy and mapping biopsies were performed. Proctoscopy was normal. Biopsies revealed extensive VIN, AIN, and well to moderately differentiated squamous cell carcinoma of the vulva.

A wide local excision of the vulva, defunctioning loop ileostomy (due to the tumour site) and reconstructive plastic surgery were preformed. Bilateral groin node dissection and ileostomy reversal were performed two months later. Lymph nodes were negative and a diagnosis of FIGO Ib vulvar SCC

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was made. Further vulvar excisions were performed at six months, and seven years later revealing recurrent VIN 3.

Twenty months post diagnosis she presented at five weeks gestation. In view of extensive vulvar scarring delivery by caesarean section was planned. Sadly at 29 weeks gestation an intra-uterine foetal death (IUFD) was diagnosed following a six-day history of decreased foetal movements. Labour was induced and a normal vaginal delivery achieved. No obvious clinical cause was identified for the IUFD, and no abnormality detected at post-mortem. At nine years following initial diagnosis the patient is alive and well with no evidence of recurrence.

DISCUSSION

Literature search revealed four prior reports,[2-5] including eleven cases of pregnancy following treatment for vulvar squamous cell carcinoma. All eleven women had prior radical vulvectomy and lymphadenectomy performed, with the majority (91%) being FIGO stage I tumours. Of these cases one antenatal complication was documented, and no incidence of disease recurrence was reported.

Including our data a total of eight women (61%) had a caesarean section; five women (38%) had a caesarean section in relation to their prior diagnosis of / or treatment for vulvar cancer; three (23%) had a caesarean section for obstetrical reasons) (Table 1).

Though it appears likely that pregnancy poses no increased risk for disease recurrence, and a prior diagnosis of vulvar carcinoma has no deleterious effects upon the antenatal period, it appears that these women may have an increased rate of delivery by caesarean section.

In recent years modifications in the surgical management of vulvar carcinoma has resulted in a reduction in surgical morbidity. Pelvic lymphadenectomy is no longer recommended,[6] triple incision techniques are more frequently employed,[7] and in early stage disease (FIGO Ia) wide local excision of the primary tumour alone is advocated as the risk of lymph node metastases in such circumstances are reported as negligible.[8] The introduction of sentinel node assessment may also result in a further reduction in lymphadenectomy for node negative disease.

As the majority of these cases (92%) were reported prior to such surgical modifications then it is possible that the incidence of delivery by caesarean section is no longer increased although vulvar scarring, plastic reconstruction, and patient choice may still influence decision making.

In successfully treated vulvar cancer subsequent pregnancy is not shown to increase the risk of disease recurrence, and there appears to be no deleterious effects during the antenatal period. It is possible, when considering prior reports, that vulvectomy may increase the likelihood of delivery by caesarean section, though modifications in the surgical management of vulvar carcinoma may have decreased this risk. It certainly seems pertinent however that those women are advised of this possibility.

The paucity of reported cases and lack of larger data series in the literature has prevented the development of accepted management plans for pregnancy following treated vulvar cancer. The authors feel that data centralisation would be beneficial in identifying optimal management strategies in not only these rare tumours, but also in other malignant tumours

Table 1. Pregnancy outcomes in women following treatment for squamous cell vulvar cancer

Reference	Age (yr)	Parity	Months post diagnosis	Mode of delivery (indication)	Gestation at delivery	Prior surgical procedure	FIGO stage	Pregnancy complications	Maternal* outcome (mon)
Gemmell[2] (1953)	37	G3P1	15	NVD	36 wk	RV+BL+VR	I	Nil	28
Dahle[3] (1959)	27	P0	36	CS (obstetric)	38 wk	RV+BIFL+PL	I	Nil	60
Rubin[4] (1960)	35	G0	24	CS (2nd look laparotomy)	Term	RV+BIPL	III	Nil	122
Collins[5] (1960)	19	G0	24	CS (vulvar scarring)	Term	RV+BL	I	Introital scarring	108
	28	G0	30	CS (vulvar scarring)	Term	RV+BL	I	Introital scarring	156
	30	G0	39	CS (obstetric)	Term	RV+BFL	I	Nil	108
	30	G3P2	24	NVD	Term	RV+BL	I	Nil	84
	32	G0	22	NVD	36 wk	RV+BL	I	Nil	132
	32	G1P1	26	NVD	Term	RV+BIL	I	Nil	72
	33	G0	32	CS (vulvar scarring)	Term	RV+BIL	I	Introital scarring	84
	39	G0	36	CS (obstetric)	36 wk	RV+BL	I	Stillbirth	120
Sheffield cases									
Case 1 (1989)	29	G2P1	24	CS (vulvar scarring)	38 wk	RV+BIL+VR	I	Introital scarring	228
Case 2 (2000)	36	P0	20	NVD	29 wk	WLE+BIL	I	IUFD	96

BIFL: bilateral inguinofemoral lymphadenectomy, BIL: bilateral inguinal lymphadenectomy, BL: bilateral lymphadenectomy, CS: caesarean section, IUFD: intrauterine foetal death, NVD: normal vaginal delivery, PL: pelvic lymphadenectomy, RV: radical vulvectomy, VR: vulvar reconstruction.

*All women in case reports were alive and well.

diagnosed and treated prior to, and during pregnancy.

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