Vulvar Carcinoma in Pregnant Women Aged Less than 40 Years: Case Report

Malihe Hasanzadeh[1], Amir Zamiri-Akhlaghi[2], Maryam Hassanpoor-Moghaddam[3], Soodabeh Shahidsales[4]

Abstract

Background: Invasive squamous cell carcinoma of the vulva is primarily a disease of postmenopausal women and thus is rarely associated with pregnancy.

Case: We have reported on a young woman under 40 years old with vulvar carcinoma, which occurred during the pregnancy but optimal treatment was delayed to the postpartum period. This 37-year-old woman was diagnosed with 3x3 cm vulvar lesion, 2 weeks after cesarean section, subsequent biopsy revealed squamous cell carcinoma. She had a history of an ulcer on her left labia minor at the third month of the pregnancy. She was treated by a modified radical vulvectomy and bilateral groin lymphadenectomy. She did not receive any additional treatments. Now after two years, she has had no recurrence of the disease.

Conclusion: This case emphasizes on the need to consider malignancy as a differential diagnosis in vulvar lesions of pregnant young women.

Keywords: Vulvar Carcinoma; Pregnancy; Squamous Cell Carcinoma

Please cite this article as: Hasanzadeh M, Zamiri Akhlaghi A, Hassanpoor Moghaddam M, Shahidsales S. Vulvar Carcinoma in Pregnant Women Aged Less Than 40 Years: Case Report. Iran J Cancer Prev. 2014; 7(3):175-8.

1. Women Health Research center, Dept. of Gynecology Oncology, Mashhad University of Medical Sciences, Mashhad, Iran
2. Dept. of Gynecology Oncology, Faculty of medicine, Mashhad University of Medical Sciences, Mashhad, Iran
3. Dept. of Gynecology Oncology, Faculty of medicine, Mashhad Azad University of Medical Sciences, Mashhad, Iran
4. Solid Tumor Treatment Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

Corresponding Author:
Soodabeh Shahidsales MD;
Assistant Professor of Radiation Oncology
Tel: (+98) 511 8461518
Email: shahidsaless@mums.ac.ir
Received: 20 Jul. 2014
Accepted: 1 Sep. 2014
Iran J Cancer Prev. 2014; 3:175-8

Introduction

Invasive squamous cell carcinoma of the vulva is primarily a disease of postmenopausal women and thus is rarely associated with pregnancy [1]. While squamous cell carcinomas account for about 90% of all primary vulvar malignancies, most squamous carcinomas of the vulva occur on the labia majora and minora (60%) [2].

Keratinizing types of vulvar carcinoma, which tend to be unifocal and occur predominantly in older patients, are not related to HPV, and often are found in areas adjacent to the lichen sclerosisand squamous hyperplasia.

Physician delay is a common problem in the diagnosis of vulvar cancer, particularly if the lesion occurs in pregnancy; therefore, any suspicious vulvar lesion detected during pregnancy should be biopsied [3]. Diagnostic delay may occur when a low suspicion of malignancy exists in the group of younger patient, due to the confusion about symptoms following the physiologic changes of pregnancy or if further investigation or treatment is postponed until the post-natal period [4].

Management is individualized [5] and with the exception of pelvic surgery, most surgical techniques that are used in non-pregnant patients are also safe for pregnant patients [6].

All patients whose tumors demonstrate more than 1 mm of stromal invasion require inguino-femoral lymphadenectomy. It is clear that it is not necessary to perform a bilateral groin dissection if the primary lesion is unilateral and the ipsilateral lymph nodes are negative [7].

Independent risk factors for recurrence include the size of the tumor, lympho-vascular space involvement, multifocality of the tumor, status of surgical margin, and the presence of a concurrent vulvar intraepithelial neoplasia [8]. In this article we presented other cases of vulvar carcinoma in the post natal period.

Case Report

A 37-year old Iranian woman (gravid 8, live 6, death 2) was referred to the Department of Gynecologic Oncology at Ghaem Hospital, in November 2009. She complained about a vulvar lesion. Two weeks before, the patient had delivered,

by cesarean section, a term infant with apgar scores of 8 and 10 at one and five minutes, respectively.

She had a history of progressive itching on her left labia minor of the vulva for 17 years and she had tried different therapies for her itching.

A punch biopsy was taken 2 years ago by a dermatologist and was diagnosedas vulvar squamous hyperplasia.

At the third month of pregnancy she noticed an ulcer on her left labia minor. She visited different physicians, but they prescribed typical ointments and antibiotics. A physical examination revealed a 3x 3 cm lesion overlying the upper labia minor (Figure 1).

She did not have palpable bilateral inguinal lymph nodes. After admission, she underwent a vulvar biopsy and her pathologic report showed a large cell keratinizing micro-invasive squamous cell carcinoma with an invasive depth of 3 mm (Figure 2).

She had a normal chest X-ray and a full sexually transmitted infection screen including HIV, syphilis serology and hemophilusducrii cultures were performed and were negative.

She was treated by a modified radical vulvectomy and bilateral groin lymphadenectomy.

The size of the invasive SCC lesion was 3×3cm with no lympho-vascularinvasion. All surgical margins of the lesion (2cm) were free. The pathologist reported a follicular hyperplasia in lymph nodes. She was discharged with no additional therapy. Two year later, she had no recurrence of the disease.

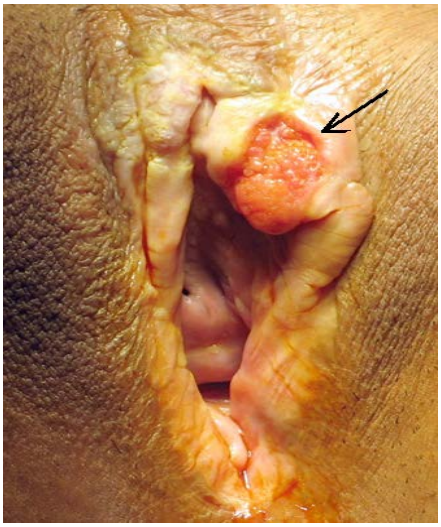

Figure 1. It shows ulcerative lesion of upper labia minor.

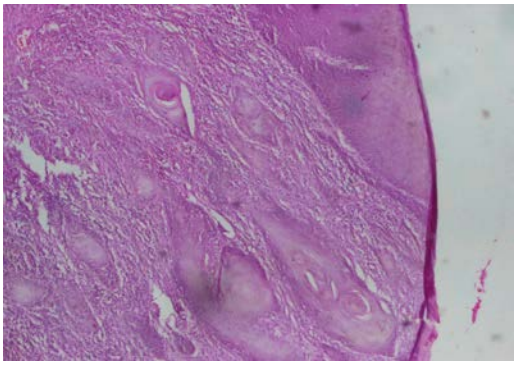

Figure 2. It shows histologic section of keratinizing micro-invasive squamous cell carcinoma.

Discussion

We have reported on a young woman under 40 years old with vulvar carcinoma, which occurred during the pregnancy. She was diagnosed during pregnancy, and so optimal treatment was delayed to the postpartum period.

Cancer of the vulva consists of 3 to 5% of all gynecologic malignancies [9], although it rarely occurs in pregnancy [1].

Up to 2010, only 26 cases have been reported as vulvar cancer during pregnancy, the latter case was reported by Keskin [10]. Some case-reports are summarized in table 1.

The most common initial symptom of vulvar cancer is pruritus, which may be of a long duration. Vulvar pain, discharge, and bleeding are less commonly reported [2]. In our patient, there was a history of pruritus for a long period.

The patient often becomes aware of a lesion on her vulva; but despite the superficial nature of the lesion, delays in seeking medical help are common. These findings underscore the need for patient and physician education with regard to the early diagnosis of carcinoma of the vulva and the importance of having a biopsy diagnosis before treating vulvar lesions.A biopsy of the vulva is a simple procedure that can be performed in the physician's office [3,4]. In our patient; there was a delay in diagnosis from the first trimester of pregnancy to puerperium.

Invasive squamous cell carcinoma of the vulva involves the labia majora in about two thirds of

Table1. Some of vulvar carcinoma case-reports in pregnancy

Title	Publisher	Case
Invasive squamous carcinoma of the vulva in women aged less than 40 years: report of two cases and a third case diagnosed during pregnancy	Eur J GynaecolOncol. 2008;29(4):399-401	In this article vulvar carcinoma was diagnosed in three women less than 40 years old one which was diagnosed in the third trimester of pregnancy. In the third patient was diagnosed during the last trimester of pregnancy and she was treated by radical surgery and postoperative radiotherapy, she had a recurrence in the inguinal at 36 months, and died of disease 12 months later
Invasive Vulvar Cancer in Pregnancy: Case Report and Current Literature Review	Journal of Lower Genital Tract Disease: 2009 V13(4) pp 264-268	The literature available to date is limited to 26 case reports.
vulvar carcinoma in pregnancy :A CASE REPORT	Medical Journal of the Islamic Republic of Iran ISSN:1016 -1430 Vol 19, Num. 2,2005, pp. 185-187	A 28-year-old Afghan woman during pregnancy presented with a vulvar squamous cell carcinoma. The patient was treated with local excision then a cesarean section in her 36th week of pregnancy. She underwent modified radical vulvectomy with bilateral inguinal lymphadenectomyfour weeks after cesarean. Because of positive groin lymph node, she also underwent radiation therapy. She is alive without invasive cancer 7 months after diagnosis.
Case Report Recurrent vulvar carcinoma in pregnancy	http://dx.doi.org/10.1016/j.ygyno.2004.07.018, How to Cite or Link Using DOI Cited by in Scopus (7)	A 36-year-old woman presented with a tender mass anterior to the left labium major, that biopsy revealed to be invasive squamous cell carcinoma. In the 23rd week of the pregnancy, she underwent a modified radical vulvectomy and bilateral inguinofemoral lymphadenectomy. Eleven weeks later, she had severe vulvar intraepithelial neoplasia (VIN III) with a small focus of invasive squamous cell carcinoma. A radical local excision was performed at 9 weeks postpartum.
Pregnancy-associated invasive squamous cell carcinoma of the vulva in a 28-year-old, HIV-negative woman. A case report	J Reprod Med, 45 (2000), pp. 659–661	The pregnant woman, HIV negative, presented with vulvar pain. She had delivered a term infant three months earlier at another institution and was diagnosed with squamous cell carcinoma of the vulva at that time but treated with delay. The patient underwent examination under anesthesia with bilateral inguinal lymph node dissection, cone biopsy, radical vulvectomy and excision of perianal lesions.
Squamous cell carcinoma of the vulva inpregnancy	Gynecol Oncol, 41 (1991), pp. 74–77	Two women presented with Vulvar carcinoma during pregnancy are reported. The first patient was treated by radical vulvectomy 2 weeks after cesarean; the second case despite underwent radical vulvectomy, died of disseminated cancer and postoperative radiation therapy. Only 12 cases of invasive squamous cell vulvar cancer during pregnancy have been previously reported.

patients [2]. Location of the lesion in our patient was in the labia minor.

Vulvar SCC in young women may occur in association with or without predisposing factors that

include age [11], cigarette smoking and venereal diseases, while chronic dermatitis is a risk factor for vulvar cancer [12].

Keratinizing types of vulvar carcinoma tend to be unifocal, and are not related to HPV, and often are found in areas adjacent to lichen sclerosis and squamous hyperplasia. Our patient had a history of chronic dermatitis for 17 years and she had a biopsy two years ago that revealed concordant squamous hyperplasia.

Because no series of a meaningful size has been reported, management is individualized [5] according to the clinical stage and depth of invasion. Our patient received standard surgical treatment and she did not require any postoperative treatment because she did not have any risk factors for recurrence, for example no lympho-vascular invasion, unifocality of the tumor, free surgical margin, and no presence of concurrent vulvar intraepithelial neoplasia.

Moreover, the single most important prognostic factor is lymph node status. This patient did not have any lymph node involvement; therefore, she did not receive any additional treatments after the surgery.

Conclusion

A careful inspection of the vulva should be a part of every gynecologic examination in pregnancy. This case emphasizes on the need to consider malignancy as a differential diagnosis in vulvar lesions of young women. We must consider biopsies for all suspicious vulvar lesions, even in young and pregnant women. Early and thorough diagnosis with subsequent appropriate definitive treatment cannot be overemphasized.

Acknowledgment

The authors would like to thank Mrs. Moshtaghi who edited this paper.

Conflict of Interest

The authors have no Conflict of interest in this article.

Authors' Contribution

Maliheh Hasanzadeh wrote the case report. Zamiri Akhlaghi and Hassanpoor Moghaddam contributed to the literature review, discussion and patient follow up. Soodabeh shahidsales contributed to write and final edit of the paper.

References

1. Palmer JE, Tidy JA. Pregnancy following vulvar squamous cell carcinoma: a report of two cases. J Gynecol Oncol. 2009; 20(4):254-6.
2. Rock JA, Thompson JD. Te lende's Operative Gynecology. 10th ed. Philadelphia: Lippincott-Raven; 2008.
3. Ghaemmaghami F, Hasanzadeh M. Good fetal outcome of pregnancies with gynecologic cancer conditions: cases and literature review. Int J Gynecol Cancer. 2006; 16(Suppl 1):225-30
4. Palmer JE, Vatish M, Tidy J. Squamous Cell Vulvar Carcinoma and Pregnancy– A Review. European Oncology. 2010; 6(2):47–52
5. Ogunleye D, Lewin SN, Huettner P, Herzog TJ. Recurrent vulvar carcinoma in pregnancy. Gynecol Oncol. 2004; 95(2):400-1.
6. Amant F, Van Calsteren K, Vergote I, Ottevanger N. Gynecologic oncology in pregnancy. Crit Rev Oncol Hematol. 2008; 67(3):187-95.
7. Berek JS. Novaks Gynecology. 14th ed. Lippincott Williams and Wilkins, 2007.
8. Preti M, Ronco G, Ghiringhello B, Micheletti L. Recurrent squamous cell carcinoma of the vulva: clinicopathologic determinants identifying low risk patients. Cancer. 2000; 88(8):1869-76.
9. 9- Sturgeon SR, Brinton LA, Devesa SS, KurmanR J. In situ and invasive vulvar cancer incidence trends (1973 to 1987). Am J Obstet Gynecol. 1992 May; 166(5):1482-5.
10. Keskin N, Iyibozkurt AC, Topuz S, Saliholu Y, Bengisu E, Berkman S. Invasive squamous carcinoma of the vulva in women aged less than 40 years: report of two cases and a third case diagnosed during pregnancy. Eur J GynaecolOncol. 2008; 29(4):399-401.
11. Bakour SH, Jaleel H, Weaver JB, Kehoe S, Radcliffe KW. Vulvar carcinoma presenting during pregnancy, associated with recurrent bone marrow hypoplasia: a case report and literature review. Gynecol Oncol. 2002 Nov; 87(2):207-9.
12. Ciszko B, Pochwałowski M, St Gabryś M. Risk factors and clinical characteristic patients with vulvar cancer. Ginekol Pol. 2006; 77(12):914-21.