Adenoid cystic carcinoma of the vulva: Conservative surgery using a de-epithelized rhomboid flap

O. Balat[1], *M.D.*; **M. Tercan**[2], *M.D.*; **M. Bekerecioğlu**[2], *M.D.*; **A. Aydın**[3], *M.D.*; **M. Karakök**[3], **İ. Kutlar**[1], *M.D.*

[1]*Department of Obstetrics and Gynecology;* [2]*Department of Plastic Surgery;* [3]*Department of Pathology*
University of Gaziantep, Sahin Bey Medical Center, Gaziantep (Turkey)

Summary

Adenoid cystic carcinoma of the vulva is a very rare entity manifested by an indolent clinical course, late recurrence and a propensity for perineural and local invasion.

A 41-year-old women underwent radical local excision and ipsilateral superficial lymph-node dissection using a de-epithelized rhomboid flap for adenoid cystic carcinoma of the left vulva. On follow-up after two years the patient is alive and well with no evidence of disease.

In this report we present the first case of adenoid cystic carcinoma of the vulva using a de-epithelized rhomboid flap.

Key words: Adenoid cystic carcinoma; Vulva; Radical local excision; Rhomboid flap.

Introduction

Adenoid cystic carcinoma (ACC) of the vulva is a very rare entity manifested by an indolent clinical course, late recurrence and a propensity for perineural and local invasion [1]. No consensus exists on adequate surgical management of this particular disease. Nevertheless, a review of the literature suggests that radical vulvectomy, wide local excision, and radical local excision should be done as the primary surgical procedure [1-3]. It also suggests that ipsilateral inguinofemoral lymph-node dissection should only be performed when suspect lymph nodes are found at clinical examination [1-3]. To the best of our knowledge, in this report we present the first case of adenoid cystic carcinoma of the vulva using a de-epithelized rhomboid flap.

Case Report

A 41-year-old women had undergone excisional biopsy for a left labial mass six months earlier. The pathologic examination revealed an adenoid cystic carcinoma (Figures 1a and 1b). At that time the patient underwent wide local excision. Thereafter, the patient was referred with a 4 to 5 cm mass originating from the left labium minus to our hospital. Physical examination revealed no palpable inguinal lymphnode. Computed tomography scan of the pelvis and abdomen revealed no pelvic-abdominal mass. The patient underwent radical wide excision and ipsilateral superficial lymph-node dissection. The excision was performed 2 cm away from the margin of the tumor. The labium minus totally, labium majus partially and one-third of the distal lateral and lower wall of the vagina were excised with the tumor. The anterior wall of the rectum and prerectal fascia was evaluated as normal. A tissue defect 12x10 cm in diameter emerged after resection of the tumor (Figure 2a). A rhomboid flap 12x10 cm in length was prepared to cover the defect (Figure 2b). The labium majus was prepared as a random pattern flap in order for the flap to reach the defected area (Figure 3a). The rhomboid flap was placed and sutured, thus some areas of the flap were de-epithelized in order to place the labial flap (Figure 3b). The normal anatomic structure was reconstructed with the flaps (Figure 4).

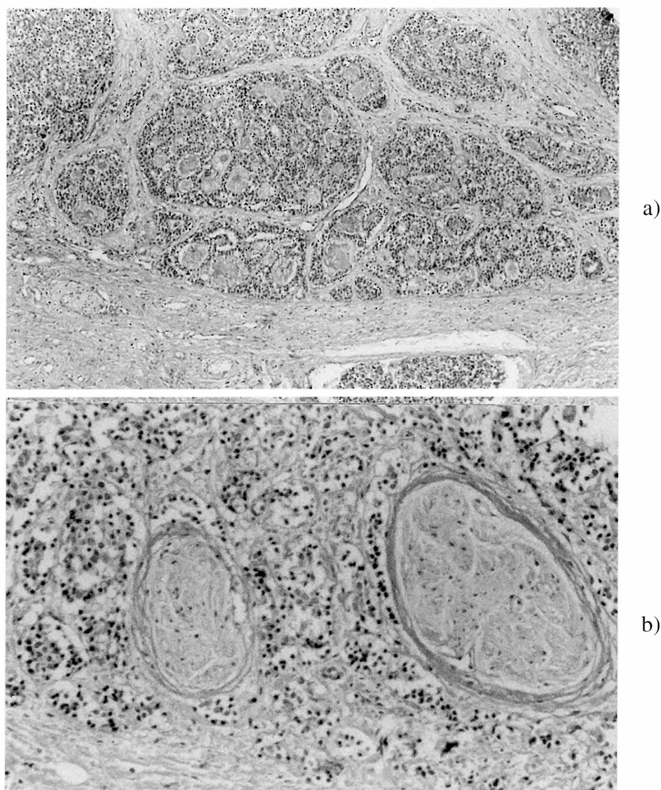

Figure 1. — **a)** Adenoid cystic carcinoma. This microphotograph illustrates basaloid tumor cells forming cribriform, tubular and trabecular structures (Hemotoxylin-eosin x 40). **b)** Perineural invasion by tumor cells is seen in this picture (H&E x 100).

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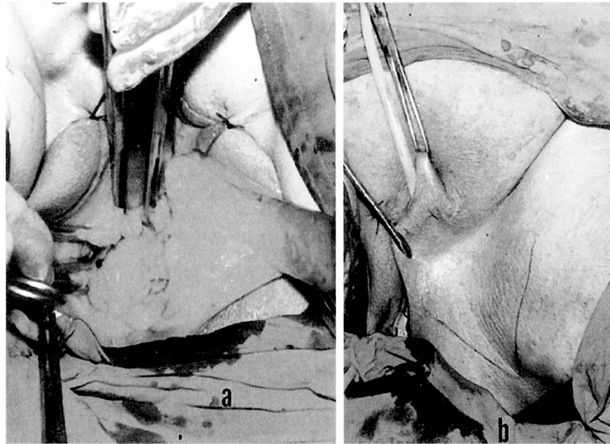

Figure 2. — **a)** Defected area after resection of the tumor. **b)** Design of rhomboid flap, inner aspect of the left thigh.

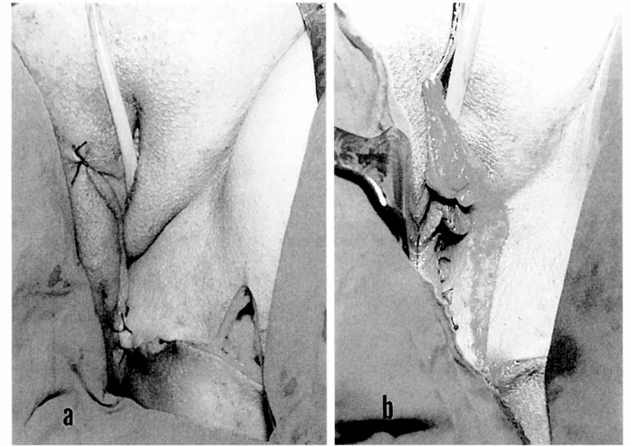

Figure 3. — **a)** Elevation of the labium majus as a random pedicle flap. **b)** De-epithelization of the rhomboid flap.

Figure 4. — Inset of the labium majus flap into the de-epithelized area and early postoperative view.

Microscopic examination revealed an adenoid cystic carcinoma with negative surgical margins. Therefore the patient received no further therapy.

Subsequent follow-up and workups have revealed no evidence of disease. Two years after initial diagnosis, she remains disease-free.

Discussion

The obvious benefits of less aggressive surgery are lower morbidity (particularly local wound break-down), shorter hospital stay, and enhanced self-image and sexual function [4]. A selective approach to dissection of the superficial groin node has been used to provide additional prognostic information [1, 2, 5].

In this case radical local excision and ipsilateral superficial inguinal lymph-node dissection were performed with negative surgical margins. There was no metastasis in all removed lymph nodes.

The local rhomboid advancement flap has proven to be an excellent option for the repair of vulvar defects not amenable to primary closure [6]. This tumor probably originates from the Bartholin's gland. Even though extensive sampling from resected material revealed a few, small, distended, benign glands nearby tumor tissue, the Bartholin's gland could not found. This situation can be explained in that the Bartholin's gland may be destroyed and wiped out by tumor tissue.

This is the first report of adenoid cystic carcinoma of the vulva in which a de-epithelized rhomboid flap was used to close a large vulvar defect after radical local excision with a very good appearance of the vulva.

There was no technique-related complication or sexual dysfunction. The patient had a high degree of acceptance of the vulvar appearance and resumed sexual function after reconstruction. On follow-up after two years the patient is alive and well with no evidence of disease.

In conclusion, we recommend radical local excision and ipsilateral superficial lymph-node dissection with a de-epithelized rhomboid flap, with less morbidity and very good vulvar appearance and sexual activity.

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Address reprint requests to:
O. BALAT, M.D.
Gaziantep University
P.T.T. Subesi, P.K. 34
Gaziantep (Turkey)