Report of two cases of controlateral groin recurrence after ipsilateral groin node dissection for vulval cancer

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Summary

Among the more conservative management strategies intended to individualize the surgical treatment of vulval cancer, ispilateral groin dissection is proposed for T1-T2 lateral lesions. Since patients found negative for metastatic nodes in the ipsilateral groin and developing unexpected recurrences in the controlateral non-dissected groin have a poor outcome, it is useful to report such cases to better evaluate the safety of these less aggressive surgical procedures.

Key words: Vulvar cancer; Groin recurrence.

Introduction

Carcinoma of the vulva accounts for 3% to 5% of all female genital malignancies. Groin lymphadenectomy is an integral part of the surgical management of invasive vulval carcinoma [1, 2]. Different conservative management strategies have been the object of trials intended to individualize the surgical approach and to reduce extensive surgery-associated morbidity: ipsilateral groin dissection is proposed for T1-T2 lateral lesions, intending to limit controlateral dissection in cases presenting positive ipsilateral nodes at frozen section analysis [3, 4]. Since patients developing unexpected recurrences in a controlateral non-dissected groin have a poor outcome, it is useful to report the occurrence of such cases to gather more information on the safety of this less aggressive procedure.

Materials and Methods

Since 1994, in an attempt to reduce the incidence of lymphoedema, seroma formation and wound breakdown, we adopted separate vulval and inguinal incisions as the standard surgical management for vulval malignancies. Groin dissection consists of inguinal and medial femoral lymphadenectomy. Cases of T1-T2 lateral lesions invading more than 1 mm up to 5 mm have been considered eligible for ipsilateral groin dissection, limiting controlateral groin dissection to cases presenting ipsilateral positive nodes at frozen section analysis [3, 5]. In such cases adjuvant radiotherapy is recommended. We report two cases of controlateral groin recurrence, after radical vulvectomy and ispilateral groin dissection for Stage II well-lateralised (medial wedge ≥ 2 cm from the midline) vulval carcinoma.

Case 1

Results

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An 81-year-old patient in 2001 underwent radical vulvectomy and ispilateral inguinal and medial femoral lymphadenectomy for Stage I, grade 2, left lateral vulval carcinoma with 5 mm of invasion. Histology of the surgical specimen only evidenced a VIN I lesion because the primary vulval lesion, measuring less than 10 mm, had been completely excised for biopsy. Nine nodes were removed, resulting negative both at frozen and definitive sections. No adjuvant therapy was given on these premises. After an uneventful 25-month period of followup, palpable right inguinal nodes were found to be metastatic at fine needle biopsy. The vulva was free from recurrent lesions. The patient underwent right inguinofemoral lymphadenectomy and four out of 17 nodes removed resulted to be metastatic.

Case 2

In 2003 an 81-year-old patient underwent radical vulvectomy with ipsilateral inguinal and medial femoral lymphadenectomy for Stage II, grade 2, right lateral invasion being (limited to 5 mm) vulval carcinoma. The primary lesion measured 40 mm. Histology of the surgical specimen showed grade 2 epidermoid carcinoma with a normal tissue margin of 1.8-2 cm. Eight nodes were removed which resulted to be negative both at frozen and definitive sections. No adjuvant therapy was done on these premises.

Six months after primary surgery, left palpable inguinal nodes were found positive for carcinomatous, grade 3, recurrence at fine needle biopsy. The patient underwent right inguino-femoral lymphadenectomy and four out of ten metastatic nodes were found.

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Discussion

In recent years vulval carcinoma has been diffusely managed through locally radical vulvectomy and separate groin incisions. Inguinal and medial femoral lymphadenectomy has become the standard approach for T1-T2 cases [5-7]. A conservative accepted approach for well-lateralized lesions up to 1 cm in diameter with invasion limited to 5 mm and no lymphovascular involvement consists of ipsilateral groin dissection, with controlateral dissection being indicated only if positive nodes are found at frozen section analysis [8, 9]. The same approach was later extended, with comparably good results, to any grade T1-T2 lateral lesions [3, 4]. Since patients developing recurrences in a controlateral nondissected groin die of their disease, such a less aggressive approach must be weighted carefully. The literature reports few cases of unexpected recurrences in a controlateral nondissected groin when ipsilateral nodes are negative for metastasis. In a GOG series six patients had controlateral groin recurrence after superficial ipsilateral groin lymph node dissection [9]. In another series two cases are mentioned of controlateral groin recurrence after complete ipsilateral inguinofemoral lymphadenectomy; in one of these cases a microscopically positive ipsilateral inguinal node was found [10]. The same occurrence is reported in one case from a NGOC series [11]. In our cases, unexpected controlateral groin failure occurred, respectively 25 and six months after primary surgery, consisting of radical vulvectomy and ipsilateral inguinal and medial femoral lymphadenectomy. Given the 8.6 ± 1.2 mean number of nodes removed in our series, eight and nine nodes were respectively removed in our two patients without evidence of nodal involvement. While aberrant lymphatic drainage to the controlateral groin may explain these unexpected recurrences [3], we emphasize, as a possible risk factor, that the primary lesion was grade 2 in both cases.

Conclusions

Given the benefit of lower morbidity, and an actuarial survival comparable to that achieved by radical vulvectomy and bilateral inguinofemoral lymphadenectomy [3, 4], controlateral groin recurrence has been considered an acceptable risk in T1 and T2 cases [3, 4]. Another option would be to perform bilateral inguinal lymphadenectomy

for neoplastic lesions more than grade 1 [11]. This would greatly reduce the chances of a less aggressive surgical approach because few cases of grade 1 are observed in T1-T2 lateral lesions with invasion of 1-5 mm and negative ipsilateral groin nodes.

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Letter to the Editor

MISDIAGNOSIS IN HEMANGIOMAS AND VASCULAR MALFORMATIONS OF THE BREAST.

Dear Editor,

With regard to Kondi-Pafitis *et al's*, recent publication "A large benign vascular neoplasm of the male breast. A case report and review of the literature" some considerations should be stated:

Accurate diagnosis of breast vascular anomalies remains a challenge for gynecologists. Tremendous confusion exists with regard to the classification and treatment of vascular lesions [2, 3]. Vascular anomalies have been categorized using inconsistent terminology and a significant number of patients receive ineffective and potentially harmful treatment based on misclassification.

In 1996, the International Society for the Study of Vascular Anomalies (ISSVA) approved a classification system in order to establish a common language for the many different medical specialists involved in the management of these lesions. Clinical, histological, histochemical, and biochemical differences and radiographic imaging findings support this classification.

A wide variety of vascular anomalies are incorrectly referred to as "hemangiomas" in the medical literature. The natural history of hemangiomas has been well documented. They proliferate at a rapid rate in the first six months of life and involute before puberty. Dissapearence occurs progressively being replaced by fibrofatty tissue by ten years of age.

The histopathology of hemangiomas is characterized by cellular markers. GLUT-1 (glucose transporter-1) is found at all phases in hemangiomas and can be extremely helpful in their differentiation from other vascular tumors [4, 5].

After the patient history and physical examination findings on the reported case we cannot support the diagnosis of *breast hemangioma*.

In conclusion, biologic classification of vascular anomalies distinguishing vascular tumors from vascular malformations is clinically useful and forms the framework for our understanding of vascular anomalies [6].

Yours sincerely,

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Authors' Reply

To the Editor,

We thank Dr. Lopez-Guttierez and Dr. Patron for their interesting comments about the classification of vascular lesions, and in particular about our reported case.

In the classification of this case we followed the strict criteria laid out in detail by Rosen in a series of articles and in his classic monograph "Pathology of the Breast" about vascular lesions of the breast.

We also applied the recent classification proposed by the World Health Organization (WHO) [2], where under the definition of breast hemangioma both neoplasms and malformations are included, without any distinction based on pathogenesis, morphology, age or sex.

It should be noted that under the chapter "Benign vascular tumors - breast hemangioma" in the same edition, the case of a 82-year-old patient with breast hemangioma is presented.

Our main interest and problem in this case was the differential diagnosis from a well differentiated angiosarcoma, which is the basic problem we encounter from a pathologist's point of view due to the extreme diagnostic difficulty well differentiated tumors present, as we comment on in our paper.

In consultation, this case was reviewed by Prof. P.P. Rosen, of the Armed Forces Institute of Pathology in the Breast and the Soft Tissue Pathology Department (case 2303092-7) where the diagnosis of breast hemangioma was made as well.

We did not apply the proposed marker GLUT-1 because it is considered as specific for juvenile hemangiomas.

We agree that the classification of breast vascular lesions must be altered to include as separate entities malformations and neoplasms, but at the time being we conform to the classification of tumors as proposed by the WHO, in order to have a common language with other pathologists and colleagues specializing in breast tumor diagnosis and treatment.

Best regards,

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Editor's Comment

I warmly thank J.C. Lopez-Gutiérrez and M. Patrón and A. Kondi-Pafitis *et al.* for their collaboration with our journal. This letter to the editor and reply allow for a deeper understanding of the problems of large benign vascular neoplasms of the breast and improve the scientific role of our journal.