

Groin recurrence following Stage IA squamous cell carcinoma of the vulva with negative nodes on superficial inguinal lymphadenectomy

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Summary

Purpose of Investigation: Stage IA vulvar cancer with a depth of stromal invasion less than 1 mm is generally managed by wide local excision alone since there is less than 1% risk of lymph node involvement. **Case:** A 62-year-old patient was admitted to a university hospital with a suspicious vulvar lesion. **Results:** We present the first case of inguinal node and a possible contralateral pubic ramus recurrence following bilateral superficial inguinal lymphadenectomy and wide local resection for Stage IA vulvar cancer. **Conclusion:** There is no evidence that extended radical surgery provides a better overall survival or reduces recurrence rate in Stage IA vulvar carcinomas. Conservative vulvar resection and sentinel node dissection seem to be a rational choice. Nevertheless the disease may recur in the inguinal areas and frequently be lethal, therefore close surveillance and early attempts to treat the recurrent disease before infection and inflammation ensues should be the aim of current treatment strategies.

Key words: Carcinoma; Groin recurrence; Inguinal lymphadenectomy; Lymphatic metastasis; Vulvar cancer; Squamous cell.

Introduction

Carcinoma of the vulva is a rare neoplasm accounting for about 5% of gynaecological malignancies [1]. The majority are squamous cell carcinoma. It primarily affects women in their sixth and seventh decades of life. Radical resection of the vulva in conjunction with bilateral groin dissection has been the standard surgical therapy for this cancer since the 1930s [2, 3]. However, this operative procedure is accompanied by substantial morbidity. Hence a more individualised treatment comprising fewer radical procedures for vulvar cancer patients has been developed in the past 30 years [4]. In 1979, DiSaia *et al.* combined some of these innovations by proposing radical local excision and superficial inguinal lymph node dissection for selected patients [5].

Stage IA tumours of the vulva and depth of stromal invasion less than 1 mm are candidates for this less radical surgery since they have less than a 1% risk of lymph node involvement [6, 7]. As a result, they are generally managed by wide local excision alone. However, eight case reports reporting lymph node metastasis in Stage IA vulvar cancer patients treated by wide local excision have been published [8-14]. In this report, we will describe inguinal lymph node metastasis in a Stage IA patient treated by wide local excision and bilateral superficial lymph node dissection.

Case Report

A 62-year-old patient presented with a 2 x 1 cm suspicious vulvar lesion on the inner aspect of the right labium majus near the clitoris. Biopsy revealed a well differentiated keratinising

squamous cell carcinoma with a depth of invasion of less than 1 mm. The tumour also had radial margin involvement. Local radical excision with bilateral superficial inguinal lymph node dissection was performed. Histopathologic evaluation of the specimen revealed no residual invasive carcinoma. There was also no evidence of metastasis in any of the harvested lymph nodes (Stage IA). No further treatment was administered.

The patient's follow-up was unremarkable for the next 27 months until she presented to the emergency clinic with a painful and infected cystic swelling of 2 cm in the right inguinal area which had begun around two months before. Immediate drainage of the cystic mass along with culture was performed and the patient was internalised for administration of systemic antibiotics. *K. pneumoniae* and *P. aeruginosa* were isolated from the lesion. Magnetic resonance imaging of the pelvis showed a 2 cm lymphadenopathy in the right groin and a 1.5 x 2 cm lesion on the acetabular joint of the left pubic ramus displaying high-signal intensity on post contrast axial T2-weighted images. Both lesions were suspicious for metastasis and inflammatory connective tissue was noted. The patient therefore underwent an excisional biopsy of the right groin lymph node, but it was incomplete due to extranodal spread of disease – especially towards the external iliac vessels – and ongoing infection. Pathology confirmed metastatic squamous cell carcinoma. Hence, her lesion in the acetabular joint was not biopsied and she was treated with palliative radiation therapy. The patient deteriorated gradually and died within four months.

Discussion

A single vulvar cancer lesion measuring 2 cm or less in diameter with a depth of stromal invasion less than 1 mm is defined as Stage IA tumour [15]. This stromal distance is measured from the base of the epithelium at the nearest most superficial dermal papilla to the deepest point of tumour penetration [16]. It has been reported that these tumours have a very low risk of lymph node involvement

(less than 1%) [6, 7], and several studies reported a risk of 0% [17, 18]. Hence, wide local excision with a tumour free margin of 1 cm without lymphadenectomy is suggested to be sufficient for treatment of these patients [19]. However, up to now, there have been reports of eight cases of regional lymph node recurrence following treatment for FIGO Stage IA squamous cell carcinoma of the vulva in the literature [8-14]. In this report, although we describe the ninth case of a patient with groin metastasis, it is the first one that developed metastasis after superficial inguinal lymph node dissection.

In our case we seemed to have overtreated our patient. Even though vulvar biopsy revealed a microinvasive carcinoma, macroscopically the lesion was around 1.5-2 cm and therefore the patient underwent lymph node dissection in addition to wide local excision due to the relative large size of the lesion. The tumour-free margin was around 2 cm and there were no metastases in all 13 harvested lymph nodes. Reexamination of the specimen confirmed a Stage 1A tumour.

It has been argued in the current literature that lymph node metastasis could have been prevented by performing superficial lymphadenectomy [13]. In Magrina *et al.* there were no groin recurrences in ten Stage 1A patients who had inguinal lymph node dissection [17]. Although superficial lymphadenectomy was performed in our patient, it did not prevent groin recurrence. This might be due to couple of reasons: if microscopic disease was present in the nodes we might have missed it. We did not perform ultra dissection of all lymph nodes dissected. It has been reported that micrometastases in superficial inguinal nodes could be missed by conventional histologic analysis [20]. Gordinier *et al.* found no micrometastases in 785 additional section cuts from negative nodes. They concluded that relapse is not a phenomenon of a false-negative pathology result [21]. Depth of invasion is the important histologic feature for predicting concurrent lymph node metastases [18]. Vulvar squamous cell carcinomas with a depth of invasion of 1.1 to 3.3 mm have an estimated risk of lymph node metastasis of approximately 10% and tumours with a depth of 3.1 to 5.0 mm have an estimated risk of lymph node involvement of approximately 15% [6].

Sentinel lymph node analysis could have helped in our situation if microscopic disease had been present in the sentinel node. Sentinel lymph node technology provides a minimally invasive surgical approach for the evaluation of inguinal lymphatic basins for metastatic disease, especially in patients with early-stage cancer. The first study regarding sentinel lymph node biopsy in vulvar cancer was published by Levenback in 1994 [22]. Terada *et al.* reported a retrospective review of 21 patients who underwent isolated sentinel lymph node dissection. With a median follow-up of 4.6 years none of these patients experienced a groin recurrence [23]. Tamussino reported a patient with micrometastasis to a sentinel node who subsequently developed a groin recurrence. They had not given any additional therapy for this micrometastasis and therefore they concluded that presence of micrometas-

tases in a sentinel node requires additional treatment [24]. Moore *et al.* found a 4.3% recurrence rate per groin or a 6.4% recurrence rate for patients that had sentinel node dissection alone, with a median follow-up of 29 months [25]. Johann *et al.* found a low false-negative rate of 2.2% [26]. Although the sentinel node biopsy procedure is determined to be a reliable procedure, it has not yet been established as a standard procedure.

If there was no disease in the resected groin nodes then microscopic disease should either have been present in the lymphatic channels between the vulvar lesion and inguinal nodes or it had metastasised to a lymph node that we did not resect: e.g. deep inguinal lymph node. In the latter case, superficial lymphadenectomy would not have been beneficial for the patient in any case.

Most groin recurrences develop earlier than vulvar recurrences, and the prognosis is much worse [27]. Lymph node status is the most powerful predictor of prognosis in vulvar cancer [28]. Groin relapse in patients who had negative nodes at superficial inguinal lymphadenectomy is uncommon, but when it occurs, it carries a very poor prognosis as in our patient [29]. Stehman *et al.* reported a 7.4% recurrence in the inguinal region after superficial inguinal lymphadenectomy [30]. Burke *et al.* reported unexpected groin relapses in 5.8% of patients with negative nodes on superficial inguinal lymphadenectomy [31]. Kirby *et al.* found an inguinal recurrence rate of 4.6% – similar to other studies evaluating superficial inguinal lymphadenectomy [32]. It is important to emphasise once again that this kind of relapse, even in patients with Stage 1A disease, has a grave prognosis. Of the reported eight cases only one patient is alive and the others died of disease. Surgery and postoperative radiotherapy was the treatment modality for the only survivor of this metastasis [14]. Unfortunately previous surgery of the inguinal area along with profuse infection and inflammation hampered our efforts of a similar type of optimal surgery for the right side. In addition, metastasis to the contralateral left pubic ramus made it impossible. This type of metastasis, as far as we know, is reported for the first time in the literature for Stage 1A patients.

Conclusion

To conclude, there is no evidence in the literature that extended radical surgery provides better overall survival, significantly improves the disease-free survival, or reduce recurrence rate in Stage 1A vulvar carcinomas. A return to radical vulvectomy with inguinal lymph node dissection may not be confirmed as it significantly increases surgical morbidity. Conservative vulvar resection and sentinel node dissection seem to be a rational choice in these patients. Nevertheless, the disease may recur in the inguinal areas and frequently be lethal. Therefore close surveillance and an early attempt to treat the recurrent disease before infection and inflammation ensues should be the aim of current treatment strategies.

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