

## CASE REPORT

# Vulval Metastasis from Squamous Cell Carcinoma of the Cervix Clinically Presenting as Lymphangioma Circumscriptum

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Patients with cervical cancer may develop local recurrence or distant metastasis, and the rate of these events is increased in proportion to the clinical stage. Cutaneous metastasis of cervical cancer is very rare and only a few cases have been reported in Korean literature. It is common at the abdominal wall, vulva, and anterior chest wall and mainly presents as an asymptomatic dermal or subcutaneous nodule, ulcer or plaque. We herein report on an interesting case of vulval metastasis from squamous cell carcinoma of the cervix with an unusual clinical manifestation resembling lymphangioma circumscriptum. (*Ann Dermatol* 23(S1) S64 ~ S67, 2011)

**-Keywords-**

Uterine cervical neoplasms, Neoplasm metastasis, Vulva

**INTRODUCTION**

Carcinoma of the cervix is a common malignancy occurring in females worldwide, and the death rate from cervical cancer ranks in the top 10 in Korea<sup>1</sup>. Patients with cervical cancer may develop local recurrence or distant

metastasis, and the rate of these events is increased in proportion to the clinical stage. Cutaneous metastasis of cervical cancer is very rare, ranging from 0.1 ~ 1.3%<sup>2</sup>, and presents mainly as an asymptomatic dermal or subcutaneous nodule, ulcer, or plaque<sup>2</sup>. We report on an interesting case of vulval metastasis from squamous cell carcinoma of the cervix with an unusual clinical manifestation resembling lymphangioma circumscriptum.

**CASE REPORT**

A 46-year-old female presented with a 3-week history of asymptomatic waxy and erythematous papules on the vulva. She was diagnosed as cervical cancer (FIGO stage Ib) 2 years ago and surgery was recommended. Due to her refusal of surgical treatment, she received treatment with concurrent chemoradiotherapy (8 cycles of cisplatin-paclitaxel chemotherapy with 41 radiotherapy treatments), and

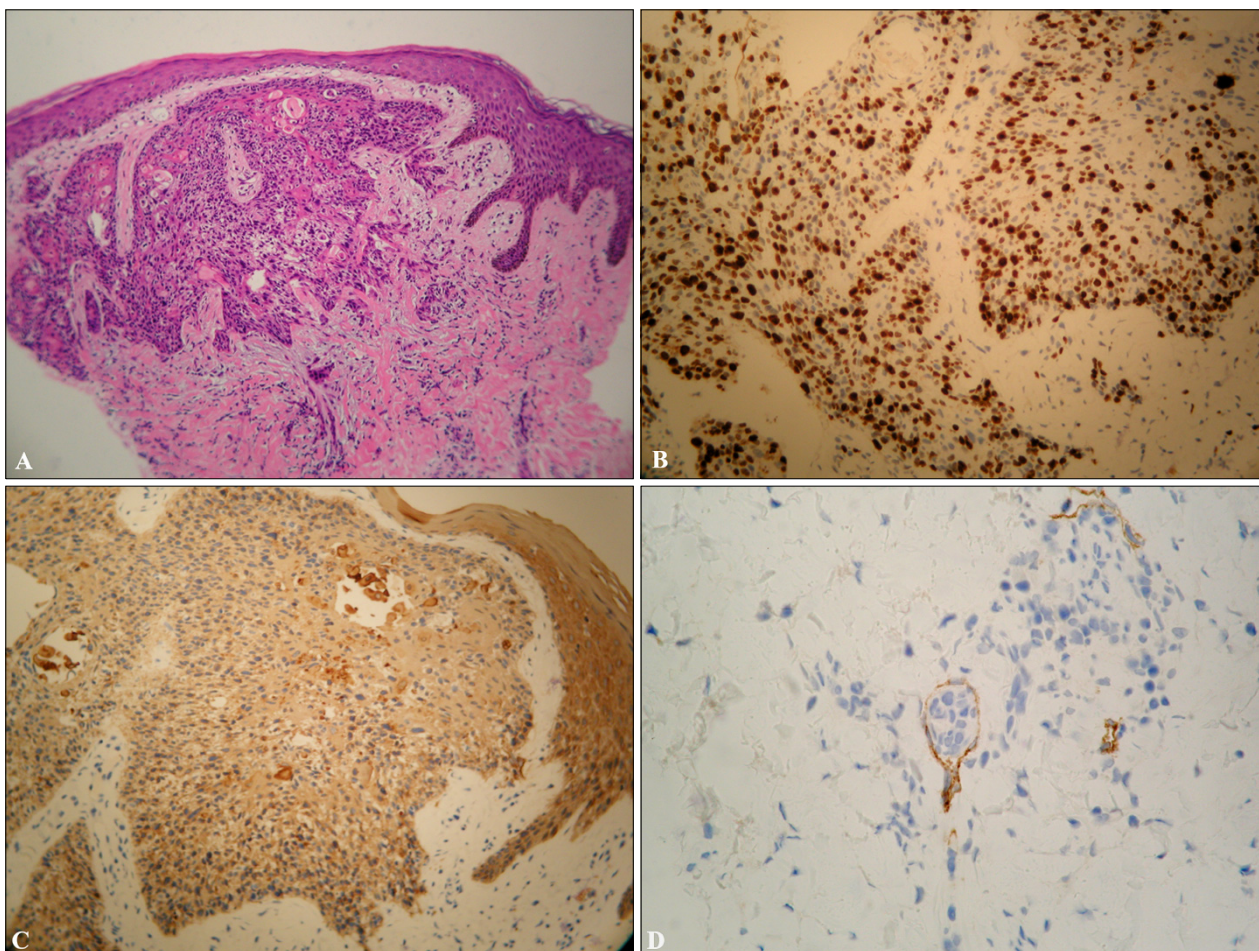


**Fig. 1.** Multiple skin-colored papules and vesicles with focal coalescence on the vulva.

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**Fig. 2.** Infiltrating tumor nests were observed in the dermis, which were composed of moderately differentiated squamous cells (A: H&E stain,  $\times 100$ ). Tumor cells were positive at Ki-67 (B:  $\times 200$ ) with a high labeling index ( $\geq 50\%$ ) and pan-cytokeratin staining (C:  $\times 200$ ) with a cytoplasmic pattern; some tumor cells were observed in the lymphatic channel (D: D2-40 stain,  $\times 400$ ).

the result was successful. However, 9 months after the end of treatment, cervical cancer recurred and she received chemotherapy again. During 4 cycles of chemotherapy, her vulval lesion appeared. On physical examination, multiple skin-colored papules and vesicles with focal coalescence on the vulva were noted (Fig. 1). Computed tomography findings of the abdomen and pelvis showed a uterine myoma measuring 3 cm and small reactive lymph nodes on the retroperitoneum; however, no specific interval change was found, compared with one year ago. Her laboratory and simple radiologic findings were also normal, except for increased squamous cell carcinoma antigen level in serum (5.01 ng/ml, reference  $< 2.0$  ng/ml). A biopsy specimen demonstrated numerous infiltrating tumor nests in the dermis, which were composed of moderately differentiated squamous cells (Fig. 2A), consistent with her primary cervical cancer. Tumor cells expressed a high Ki-67 labeling index ( $\geq 50\%$ ) and showed positivity on pan-cytokeratin staining with a cyto-

plasmic pattern (Fig. 2B, C); tumor cells were also observed in the lymphatic channel (Fig. 2D). She underwent a vulvectomy and received 6 cycles of cisplatin-paclitaxel chemotherapy, and she has remained alive for 6 months after the initial diagnosis of cutaneous metastasis.

## DISCUSSION

Cutaneous metastasis of cervical cancer is common at the abdominal wall, vulva, and anterior chest wall<sup>2</sup>; however, cases presenting at the scalp<sup>3,4</sup>, extremities<sup>5</sup>, umbilical surgical scar<sup>6,7</sup>, and drainage site<sup>8</sup> have been reported. Only a few cases have been previously reported in Korean literature (Table 1)<sup>9-11</sup>. Most lesions present as single or multiple asymptomatic subcutaneous nodules, plaque, ulcer, or inflammatory telangiectatic areas<sup>12</sup>. However, our patient had a very unusual clinical feature, like that of lymphangioma circumscriptum.

Lymphangioma circumscriptum is a rare benign disorder of

**Table 1.** Cases of cutaneous metastasis of cervical cancer reported in Korean literatures

Reference	Age	FIGO stage	Initial treatment	D/M interval (months)	Sites	Clinical manifestation	Treatment of metastasis	Outcome (months)
Park et al. (2000) <sup>9</sup>	57	IIA	RTx → recur → CTx	131	Vulva	Ulcerative nodules	RTx	Expired (12)
	46	IIB	CTx	22	Rt.thigh	Erythematous nodules and ulcer	RTx	Expired (6)
Kim et al. (2000) <sup>10</sup>	58	IIA	TAH&BSO → recur → RTx	68	Rt.thigh	Erythematous to brownish indurated papules and nodules	RTx	Expired (8)
Lee et al. (2009) <sup>11</sup>	56	IIA	CTx+TAH&BSO → RTx	8	Vulva	Erythematous ulcerative papules and nodules	CTx	Follow up
Kim et al. (our case)	46	Ib	CTx+RTx → recur → CTx	24	Vulva	Erythematous grouped papules and vesicles	Surgery +CTx	Follow up

D/M interval: interval between diagnosis of cervical cancer and that of cutaneous metastasis, CTx: chemotherapy, RTx: radiotherapy, TAH&BSO: total abdominal hysterectomy and bilateral salpingo-oophorectomy.

lymphatic channels in dermis characterized by the appearance of multiple small and discrete skin-colored vesicles<sup>13</sup>. Histopathologically, numerous dilated lymphatics are lined by a single layer of endothelial cells in the papillary dermis. Occurrence of lymphangioma circumscriptum of the vulva is relatively rare and the most common predisposing condition is surgery and/or radiotherapy<sup>13,14</sup>. We first supposed lymphangioma circumscriptum due to her history of radiotherapy and complete remission after previous treatment. However, because the lesions were solid on palpation, cutaneous metastasis had to be differentiated. Her biopsy specimen showed numerous infiltrating tumor nests in the dermis, which were composed of moderately differentiated squamous cells; based on the experience of this case, not only lymphangioma circumscriptum, but also cutaneous metastasis, should be considered when the vulval lesion resembles lymphangioma circumscriptum. At onset, lymphangioma circumscriptum commonly manifests as small millimeter-sized soft clear vesicles, whereas metastatic lesions are relatively firm and often appear in papulonodular structures. Therefore, careful history taking and palpation are important in differential diagnosis.

Cutaneous spread of primary cancer occurs by direct extension, or dissemination via bloodstream or by lymphatics. In cervical cancer, tumor cells metastasize through retrograde lymphatic dissemination secondary to lymphatic obstruction<sup>2,15</sup>. Our case supports this idea that the patient showed clinical evidence of lymphatic obstruction and leg edema after radiotherapy, and the histopathology showed cancer cells in the dilated lymphatic channel.

Although skin metastasis of cervical cancer is thought to indicate a 'pre-terminal', or 'end-stage' disease, some patients survived longer than a year with treatment of a combination of surgical excision and chemotherapy or

radiotherapy<sup>2,5</sup>. Our patient has also survived longer than 6 months by treatment with surgery and chemotherapy.

In summary, this is a very interesting case in that cutaneous metastasis of cervical cancer clinically resembled lymphangioma circumscriptum. In the case of multiple small clear vesicles on the vulva, lymphangioma circumscriptum is the foremost impression; however, cutaneous metastasis, particularly in the cervix of cancer patients, should be included in differential diagnosis.

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