Acquired Vulvar Lymphangiomas: A Sequela of Radiation Therapy

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Lymphangiomas of the vulva are rare clinical entities. Acquired or secondary lymphangiomas have characteristically been reported after radiation therapy for cervical carcinoma and appear on the vulva years after this treatment. Local surgery, scrofuloderma, and Crohn’s disease may also damage vulvar lymphatic flow and lead to the development of vulvar lymphangiomas. We report a case of acquired vulvar lymphangiomas that occurred in a patient 15 years after she received radiation therapy for squamous cell carcinoma of the uterine cervix.

Acquired or secondary lymphangiomas of the vulva are rare clinical entities that represent dilated, ectatic, superficial dermal lymphatic channels. The term lymphangiectases also is used to describe acquired lymphangiomas. Vulvar lymphangiomas may occur idiopathically or commonly after injury to vulvar lymphatic flow resulting from local surgery, scrofuloderma, or Crohn’s disease. Characteristically, a history of radiation therapy for cervical carcinoma has been the documented cause of lymphatic insult and obstruction. A case of acquired vulvar lymphangiomas is reported that occurred in a patient 15 years after she received radiation therapy for squamous cell carcinoma of the uterine cervix.

Case Report
A 79-year-old woman presented with a 1-year history of recurrent weeping and oozing from the labia, which required that she wear absorbent pads. She experienced occasional blister formation and labial burning, but denied any itching, pain, or vaginal discharge. Self-treatment with an over-the-counter vaginal cream containing benzocaine and resorcinol was ongoing. Fifteen years prior, the patient underwent radiation therapy for the treatment of squamous cell carcinoma of the uterine cervix.

On examination, flesh-colored to erythematous, edematous, boggy, oozing plaques were present on the labia bilaterally. Within these plaques, 1- to 2-mm translucent vesicles were scattered and appeared both singly and in groups (Figure 1). A Tzanck test and viral cultures were negative. Discontinuation of the over-the-counter vaginal cream and a short course of treatment with a mid potency topical corticosteroid cream had no effect on the cutaneous disorder. A punch biopsy revealed dilated channels in the papillary dermis lined by a single layer of flat endothelial cells (Figure 2). These channels were filled with an eosinophilic, homogenous material. Epidermal hyperkeratosis and acanthosis also were present.

Comment
Acquired vulvar lymphangiomas, or lymphangiectases, is a condition in which superficial dermal lymphatic channels become dilated and ectatic. Obstruction to previously normal lymphatic flow is the recognized pathologic event that leads to clinically apparent lesions. Vulvar lymphatic
obstruction may occur after local surgery, or it may appear in conjunction with scrofuloderma or Crohn’s disease. Additionally, a history of radiation therapy for the treatment of cervical carcinoma is recognized as a cause in numerous cases in which skin lesions appear years after the radiation insult.

Clinically, acquired lymphangiomas on the vulva may appear as single or grouped, translucent to blood-tinted vesicles ranging in size from minute to 5 mm in diameter. The lesions have been described as resembling frog spawn and may be mistaken for genital warts, herpetic vesicles, hemangiomas, filariasis, lymphogranuloma venereum, tuberculosis verrucosa cutis, and contact dermatitis, as in our case. Acquired lymphangiomas may occur either with or without underlying vulvar edema. Symptomatically, patients commonly complain of ruptured vesicles that weep and ooze, a leakage that is representative of lymphorrhea. Additional symptoms may include vulvar burning, itching, pain, bleeding, and sexual dysfunction. Furthermore, vulvar lymphangiomas may act as portals of entry for infection, which can lead to recurrent vulvar cellulitis.

Biopsy of the vesicular lesions is diagnostic, revealing dilated lymphatic channels in the papillary dermis lined by a single layer of flat endothelial cells. Variable amounts of eosinophilic, homogenous material that represents lymphatic fluid or red blood cells may be present. In addition, overlying epidermal hyperkeratosis and acanthosis commonly occur. Histologic features of a radiation dermatitis are not typically seen, despite the role of radiation in causing lymphatic obstruction. Malignant transformation has not been reported to occur in acquired vulvar lymphangiomas; however, chronic lymphedematous areas must be under continual surveillance for the possible development of lymphangiosarcoma. Lymphangiomalike lesions occurring on the buttocks and thigh of a patient, representing cutaneous metastases from carcinoma of the cervix, have been reported.

Treatment modalities utilized for vulvar lymphangiomas have included carbon dioxide laser vaporization, excisional surgery, ‘electrosurgery,’ cryotherapy, and sclerotherapy. The best consistent results have occurred with carbon dioxide laser vaporization and excisional surgery. However, recurrences have been reported.

In summary, acquired lymphangiomas of the vulva may present after a variety of insults to vulvar lymphatic flow. A history of radiation therapy for cervical carcinoma is now a well-recognized association.

REFERENCES