Cryotherapy for Massive Vulvar Lymphatic Leakage Complicated with Lymphangiomas Following Gynecological Cancer Treatment

Shintaro Yanazume1,*, Haruhiko Douzono2, Hidemichi Kubo3, Tomomi Nagata1, Tsutomu Douchi1 and Hiroaki Kobayashi1

1Department of Obstetrics and Gynecology, Faculty of Medicine, Kagoshima University, Kagoshima, 2Dozono Medical House, Kagoshima and 3Department of Dermatology, Faculty of Medicine, Kagoshima University, Kagoshima, Japan

*For reprints and all correspondence: Shintaro Yanazume, MD, Department of Obstetrics and Gynecology, National Hospital Organization Kagoshima Medical Center, 8-1 Shiroyama, Kagoshima 892-0853, Japan.
E-mail: s-yana@kagomc2.hosp.go.jp

Received April 16, 2014; accepted August 2, 2014

Vulvar lymphatic leakage is a severe complication associated with gynecological cancer treatments. However, standard treatment strategies have not yet been determined. We encountered a rare case of a 76-year-old multiparous woman suffering from massive lymphatic fluid leakage from the entire vulva, and papules developed and were identified as lymphangiomas. A large amount of straw-colored discharge continued from all vulvar papules, which extended over the mons pubis. Nine years ago, the patient had undergone a radical hysterectomy with concurrent chemoradiation for uterine cervical cancer treatment. Her serum albumin level was 1.9 mg/dl, which was attributed to the loss of a large amount of lymph fluid due to leakage from the vulva. Her quality of life gradually decreased because of general fatigue and the need for frequent diaper exchanges every 2 h. The patient received a less-invasive treatment with cryotherapy using liquid nitrogen. She also received a multimodality treatment consisting of the intravenous administration of albumin, massage of the lower limbs and intensive rehabilitation. Cryotherapy was administered once a week for 3 months. Her discharge almost stopped and vulvar lymphangiomas decreased without any major complications. To the best of our knowledge, this is the first case report of massive lymphatic leakage complicated with vulvar lymphangiomas. Additionally, this case may represent the first successful treatment of vulva lymph leakage by cryotherapy without recurrence. Cryotherapy may have the potential to improve the quality of life as a less-invasive treatment for gynecological cancer survivors without serious complications.

Key words: cryotherapy – vulva – lymphangioma – lymphatic leakage

INTRODUCTION

Lymph leg edema is the most common and serious manifestation in patients with gynecological malignancies. Lymph edema triggers include damage to the lymphatic vessels after surgery, chronic infections, alterations in connective tissue caused by radiotherapy and obstruction of the lymphatic vessel (1). Advanced lymph edema can cause subcutaneous lymphatic cisterns, which have lost their drainage capacity in the deep lymphatic network, and the leakage of lymph fluid over the skin (2). Acquired lymphangiomas are rare benign proliferations of the lymphatic system, the fundamental and underlying etiology is thought to be chronic lymphedema due to impaired lymph flow (3, 4).

Previous studies described acquired vulvar lymphangiomas following gynecological cancer treatment (3–5). The treatment strategies examined included excision by vulvectomy and laser vaporization for invasive treatment. Cryotherapy was described as a less-invasive treatment option for vulvar lymphangioma. However, the efficacy of cryotherapy in
patients with vulvar lymphatic leakage has not been confirmed. This case report describes the successful treatment of massive vulvar lymphatic leakage complicated with lymphangiomata with cryotherapy in a uterine cervical cancer survivor.

CASE

Full informed consent was obtained from the patient. This study was conducted in accordance to the Declaration of Helsinki (as revised in Tokyo 2004). A 76-year-old, multiparous woman was referred to our hospital with massive vulvar lymph fluid leakage complicated with bilateral lymph leg edema. Her height and body weight were 154 cm and 48 kg, respectively. In 2004, the patient underwent a radical hysterectomy with concurrent chemoradiation (pelvic radiation and intra-cavitary radiation) for FIGO Stage Ib squamous cell carcinoma of the uterine cervix. However, she developed bilateral lymph leg edema in 2007, and was being treated with compression stockings and massage. She also had massive lymph fluid leakage from the entire vulva, which caused papules in the vulva. She was intravenously administered albumin weekly on a regular basis by a local practitioner. In spite of this treatment, her quality of life gradually decreased because of general fatigue and the need for frequent diaper exchanges every 2 h. She recently lost the ability to walk unassisted.

In our outpatient clinic, pressure was applied to the vulvar tumor using large gauze; however, discharge persisted beyond the capacity of the gauze. A large amount of straw-colored discharge continued from all the vulvar papules, which extended over the mons pubis (Fig. 1). The patient had dementia, and appeared to be in a state of depression. Her serum hemoglobin and albumin levels were 14.5 g/dl and 1.9 mg/dl, respectively. The lower serum albumin levels were attributed to the loss of a large amount of lymph fluid due to leakage from the vulva. The pathological findings of vulvar tumor biopsy revealed lymphangiomas (Fig. 2), but no evidence of cancer recurrence based on imaging and a local examination. Wide excision was initially considered for vulvar lymphangiomas, and surgical intervention was discussed with our dermatology/plastic surgery department. The patient could not walk by herself and risked of developing severe complications from performing invasive surgery. Therefore, she received the less-invasive treatment of hyperbaric gaseous cryotherapy using liquid nitrogen for all skin lesions. She also received a multi-modality treatment consisting of the intravenous administration of albumin, massage of the lower limbs and intensive rehabilitation. Cryotherapy was performed once a week for 3 months. No side effects were reported, except for pain that did not require opioids. The surface of the tumor eventually became flat and vulvar lymphangiomas decreased (Fig. 3). Her discharge almost stopped, and she regained mobility due to the marked improvement in her leg edema. There has been no further vulvar tumor or discharge 8 months after these treatments.

COMMENT

Patients with vulvar lymphatic leakage due to the progression of lymph leg edema following radical hysterectomy are rarely encountered. Therefore, the management of vulvar lymph leakage has not been standardized. Reported cases of acquired lymphangioma caused by impaired lymph flow have been increasing recently. Approximately 60 cases of acquired lymphangiomas were obtained from a MEDLINE search (search terms: Vulvar or Vulva, lymphangiomas) and research review (6–8). The review revealed that 55% of acquired lymphangioma patients previously underwent radical surgery and/or radiation as cancer treatments (3). Genital tuberculosis and Crohn’s disease also commonly causes this condition.

The traditional treatment comprising the surgical removal of lymphangiomas is frequently unsuccessful (4, 6). Two-thirds of the reported cases of lymphangiomas underwent surgical treatment (7). However, the recurrence rate following excision was 25% with follow-up periods ranging from 6–81 months (3). The extent of lesions at presentation has also been shown to affect surgical outcomes and recurrence. Older patients are more likely to be unable to undergo surgical excision because of acquired lymphangiomas. Some elderly patients, including this patient described herein also had severe complications or a status with palliative sessions (3). Vaporization with a carbon dioxide laser has been used as an effective treatment for lymphangiomas (4, 8, 9). However, treatment failure was reported in long-term outcomes, as well as the new formation of keloids (4).

Cryotherapy involves the application of any substance to the body that removes heat, resulting in a decreased tissue
temperature. Topical cold treatments decrease the temperature of the skin to a tissue depth of 2–4 cm. The cryotherapy technique applies a direct hyperbaric liquid nitrogen spray onto the surface of the vulva for \(\approx 20\) s depending on the size of the tumor. Cryotherapy decreases tissue blood flow by vasoconstriction and reducing tissue metabolism and inflammation (10). The complications associated with cryotherapy include superficial nerve neuropathy, cardiovascular side effects, Raynaud’s phenomenon and frostbite (10). Only a small number of patients have received cryotherapy for vulvar lymphangioma, which appeared to be ineffective until this case report (4, 11). Recently, lymphaticovenular anastomosis has been widely performed to reduce lymphedema after gynecologic cancer treatment as a less-invasive treatment. Although cryotherapy is mainly used in treating the surface of the skin, lymphaticovenular anastomosis affects deeper vessels. Thus, a combination of cryotherapy and lymphaticovenular anastomosis may have the potential to be more effective for vulvar lymph leakage than cryotherapy alone in other gynecologic cancer survivors.

In our case, a massive amount of lymph fluid leaked from the surface of all lymphangiomas, but the patient refused invasive treatment. Cryotherapy could be used to stop vulvar lymph leakage resulting from lymphangiomas. Almost complete remission could be achieved by combining cryotherapy with a multimodality treatment. The benefits of cryotherapy continued for 8 months. No side effects were reported, except for slight pain that did not require opioids. The reason why our patient was successful with cryotherapy may have been due to the atypically extended duration of the treatment. Continuous coolant spray decreased lymph flow and improved the tissue environment. The successful treatment of lymphangioma may have consequently reduced vulvar lymphatic leakage. To the best of our knowledge, this is the first case report of massive lymphatic leakage complicated with vulvar lymphangioma. This case may also represent the first successful treatment of vulvar lymphatic leakage by cryotherapy without recurrence. Cryotherapy can be easily applied in outpatient clinics because it is commonly used in dermatology.

In conclusion, cryotherapy may have the potential to improve the quality of life of patients with lymphatic leakage as a less-invasive treatment for gynecological cancer survivors without serious complications. However, we have only encountered a small number of subjects, and there is currently no information on the use of cryotherapy for an extended period of time. Therefore, further extensive studies are necessary and warranted.

**Conflict of interest statement**

None declared.

**References**


