A case of cellulitis that complicated lymphedema of the lower limb and produced systemic inflammatory response syndrome (SIRS)

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Summary

This is the first report describing lymphedema of the lower limb after surgery for ovarian cancer complicated by severe cellulitis with systemic inflammatory response syndrome (SIRS). Debridement of the lesion and a split-thickness graft were required for a complete cure. Although lymphedema as a complication after surgery for gynecologic malignancies is prone to be considered less serious on the medical side, it is considered necessary to treat lymphedema of the lower limbs while it is still in a mild state since severe cellulitis may accompany lymphedema, as in the present case.

Key words: Lymphedema; Cellulitis; Systemic inflammatory response syndrome; Ovarian cancer; Lymphadenectomy; Split-thickness graft.

Introduction

Lymphedema of the lower limbs is one of the complications that may develop after a pelvic lymphadenectomy is performed as part of the surgical treatment for gynecologic malignancy. Lymphedema, once developed, rarely heals without appropriate treatment. It often causes inflammation, as well as cosmetic problems and movement limitation. It is also often complicated by cellulitis, lowering the patient's quality of life.

We report a case of cellulitis that accompanied lymphedema of the lower limb which developed after ovarian cancer surgery and which produced systematic inflammatory response syndrome (SIRS).

Case report

The patient was a 63-year-old woman who underwent ovarian cancer surgery in May 2000. She also underwent postoperative anticancer chemotherapy for Stage IIIc ovarian cancer. She had been visiting the hospital as an outpatient without any signs of recurrence. Lymphedema of the right lower limb had been observed since early 2002. In August 2003, severe pain in the plantar region to the dorsum of the right foot suddenly developed and she was admitted to our department. She was in shock on admission with a body temperature at 35.6°C, blood pressure of 60/30 mmHg, and pulse of 120/min. She had an agonized look due to pain in the lower limb. Her right lower leg showed edematous swelling and reddening. It was partly dark red from the plantar region to the dorsum of the right lower limb, and she could hardly walk due to the pain. Hematological examination showed a WBC count of 1.170/mm^3, Hgb of 12.7 g/dl, and platelet count of 92.000/mm^3. CRP was elevated to 27.1 mg/dl. Blood culture was negative. D-glucan in the venous blood was also negative. Hemolytic streptococcal infection was also negative. A CT scan ruled out deep vein thrombosis and pulmonary embolism. Infection complicating lymphedema of the lower limb was suspected from the symptoms and laboratory findings on admission. The patient was in a state of circulatory failure. In addition, she was considered to have developed SIRS due to infection as her clinical state satisfied three criteria (body temperature ≤ 36°C, heart rate ≥ 90/min, WBC count < 4.000/mm^3) [1]. Directly after admission, antishock and anti-inflammatory therapy was started with fluid replacement, glucocorticoids, urinastatin, dopamine hydrochloride, and antibiotics. From the second day of admission, a fever of more than 39°C was noted, the dark-reddish part of the dorsum of the foot broadened, and blistering started to appear sporadically. Although the causative organisms were not identified, the patient was diagnosed with cellulitis that complicated lymphedema of the lower limb. In the portion with blistering from the dorsum to the plantar region of the right foot, excoriation started, ulceration was recognized (Figure 1A), and multiple abscesses were drained from the skin ulcer site over time. As necrosis was noted in the lateral dorsum of the right foot, the necrotic region was excised and pus was drained by incision (Figure 1B). However, as the abscess formation in the dorsum of the right foot was worsening, debridement and a split-thickness skin graft were performed one month after the onset in order to remove the source of infection. The debridement was performed to the surface of the fascia, and the subcutaneous tissue with trapped pus that constituted the main body of the inflammation was eradicated (Figure 2A) and fixed as a mesh skin graft (Figure 2B). As the fever went down four days after the surgery and CRP decreased to within normal range, the treatment with antibiotics was terminated on the seventh day after surgery. The patient was able to sit two weeks after the surgery and walk one month after the surgery. Figure 3 shows the recovering process of the right lower limb from cellulitis two months after surgery.

Discussion

We often experience a case in which lymphedema of the lower limbs develops after surgery for a gynecologic malignancy, with recurrent infection. However, this is the first case to be reported in which lymphedema of the

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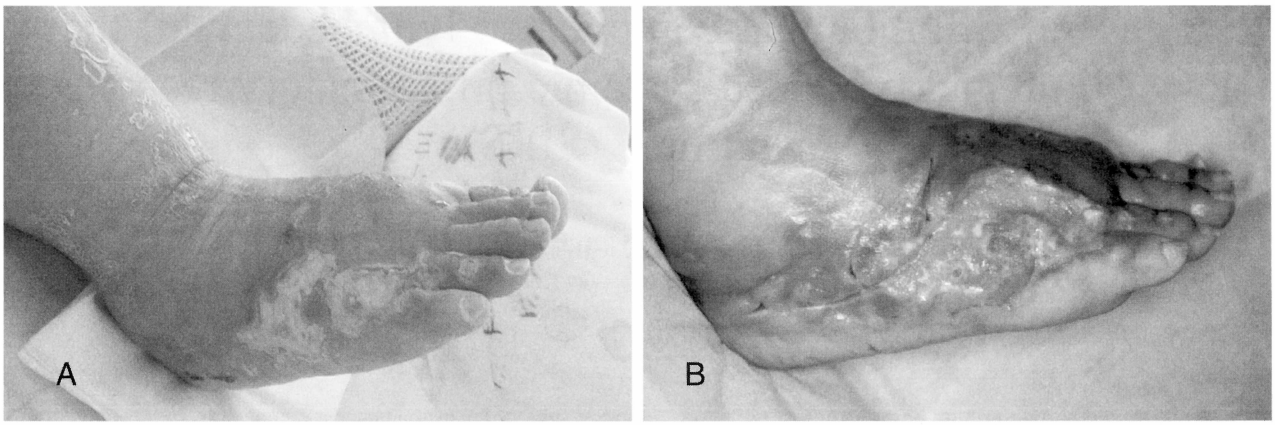

Figure 1. — Lymphedema of the right lower limb complicated by severe cellulitis. A: Blistering, excoriation and ulceration from the dorsum to the plantar of the right foot. B: Necrosis in the dorsum of the right foot. The necrotic region was excised and pus was drained by incision.

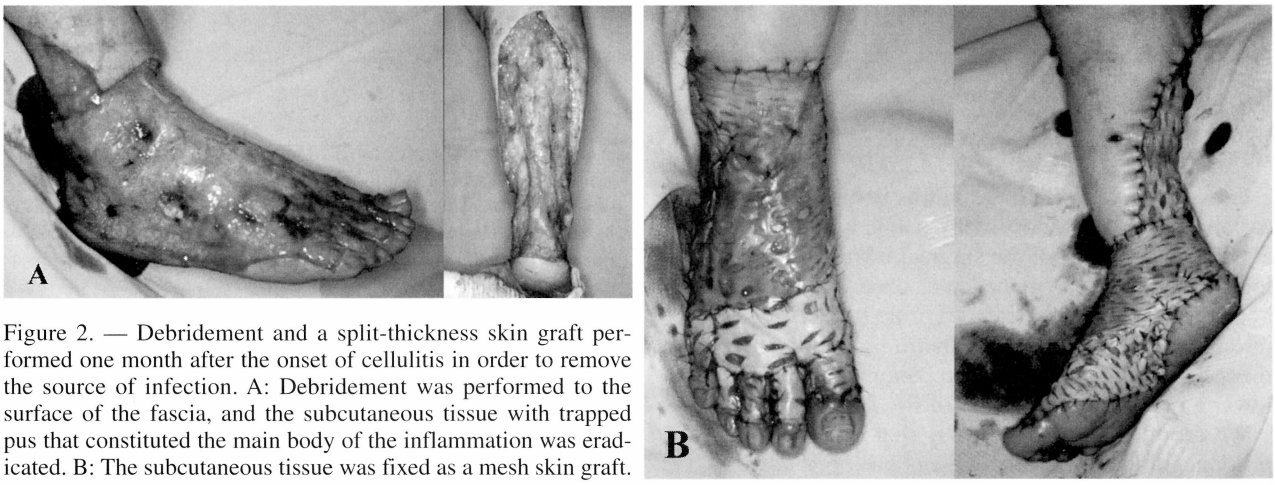

Figure 2. — Debridement and a split-thickness skin graft performed one month after the onset of cellulitis in order to remove the source of infection. A: Debridement was performed to the surface of the fascia, and the subcutaneous tissue with trapped pus that constituted the main body of the inflammation was eradicated. B: The subcutaneous tissue was fixed as a mesh skin graft.

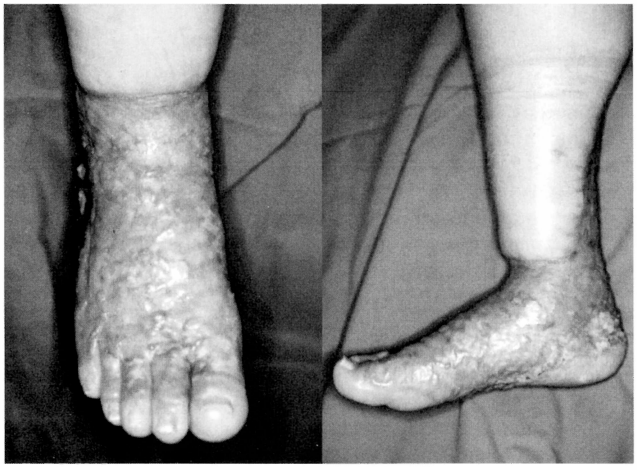

Figure 3. — Recovering process of the right lower limb two months after surgery.

lower limb after surgery for ovarian cancer was complicated by severe cellulitis with SIRS and even debridement of the lesion and a split-thickness graft were required for a complete cure.

Lymphedema of the lower limbs is said to develop due to the stagnation of the lymph fluid that occurs when lymphatic vessels are blocked in the pelvic cavity or deep-seated lymphatic return from the lower limbs is interrupted by pelvic lymphadenectomy or by the pressure of recurrent tumor. There is a report that addition of postoperative radiation therapy increases the risk of lymphedema of the lower limbs [2]. This is considered to be due to the formation of collateral vessels by the superficial lymphatic tract which is disturbed, thus resulting in insufficient compensation. Preventive measures against lymphedema of the lower limbs after pelvic lymphadenectomy include a method in which an operation is finished without suturing the pelvic peritoneum [3] or a method in which pelvic lymphadenectomy itself is avoided by performing sentinel-lymph node biopsy [4]. However, the efficacy of either method has not been established.

Lymphedema often induces recurrent inflammation, causing fibrosis of lymphatic vessels and their surrounding tissue that have carried out compensatory function. A vicious cycle is formed when lymphatic return decreases to worsen the lymphedema and make inflammation easier

to develop [5]. Cellulitis is generally considered as an infectious, inflammatory disease, and β hemolytic streptococcus is believed to be the most probable pathogenic bacteria [6]. However, it is not clear if cellulitis is caused by the bacterial product or bacterial antigen, while there is also a theory that a fungal antigen may be responsible [6], leaving much to be elucidated with regard to the causative substance. Even stranger, despite the dominant theory that cellulitis is a bacterial infection, is that the detection rate of bacteria from the lesion is never high. The reality is that the detection rate is no more than 20% in skin biopsies and bacteria cultures according to the report by Hook *et al.* [7] and 5-26% according to other reports [8]. In the present case, causative organisms were not identified, and both hemolytic streptococci and fungi were negative.

A noteworthy theory claims that the development of cellulitis is related to environmental factors generated in the region where lymphedema has developed. Mallon *et al.* described that because lymphatic return is markedly reduced between peripheral tissue and the regional lymph node, the immunity of regional extremities is lowered, and lowered immunity in the region easily induces and exacerbates infection [9].

Lymphedema is a common complication after pelvic lymphadenectomy, but from a medical perspective it is prone to be considered less serious, as its sole existence does not directly affect cancer treatment. However, besides cosmetic problems or motor deterioration, severe cellulitis accompanies lymphedema on rare occasions as in the present case. Thus, it is necessary to actively follow lymphedema of the lower limbs even while it is still in a mild state. The treatment of lymphedema includes surgical therapies such as excision and lymphatic induction as well as conservative therapy. However, an effective therapy has not been established, and complex physical therapy that combines the use of prophylactic elastic stockings, lymphatic massage, and skin care remains the center of the treatment [10]. Therefore, in high-risk cases of lymphedema where pelvic lymphadenectomy has been performed, it is necessary to provide education and lifestyle guidance about lymphedema even before its development.

References

[1] Ahmed N.A., Yee J., Giannias B., Kapadia B., Christou N.V.: "Expression of human neutrophil L-selectin during the systemic inflammatory response syndrome is partly mediated by tumor factor alpha". *Arch. Surg.*, 1996, *131,* 31.

[2] Soisson A.P., Soper J.T., Clarke-Pearson D.L., Berchuck A., Montana G., Creasman W.T.: "Adjuvant radiotherapy following radical hysterectomy for patients with stage IB and IIA cervical cancer". *Gynecol. Oncol.*, 1990, *37,* 390.

[3] Srisomboon J., Phongnarisorn C., Suprasert P., Cheewakriangkrai C., Siriaree S., Charoenkwan K.: "A prospective randomized study comparing retroperitoneal drainage with no drainage and no peritonization following radical hysterectomy and pelvic lymphadenectomy for invasive cervical cancer". *J. Obstet. Gynaecol. Res.*, 2002, *28,* 149.

[4] Martinez-Palones J.M., Gil-Moreno A., Perez-Benavente M.A., Roca I., Xercavins J.: "Intraoperative sentinel node identification in early stage cervical cancer using a combination of radiolabeled albumin injection and isosulfan blue dye injection". *Gynecol. Oncol.,* 2004, *92,* 845.

[5] Kariya M.: "Lymphedema of the lower extremeties after surgical treatment for gynecologic malignant tumors (in Japanese)". *Adv. Obstet. Gynecol.,* 2003, *55,* 144.

[6] Baddour L.M., Bisno A.L.: "Non-group A beta-hemolytic streptococcal cellulites. Association with venous and lymphatic compromise". *Am. J. Med.*, 1984, *76,* 172.

[7] Hook E.W.3rd., Hooton T.M., Horton C.A., Coyle M.B., Ramsey P.G., Turck M.: "Microbiologic evaluation of cutaneous cellulitis in adults". *Arch. Intern. Med.*, 1986, *146,* 295.

[8] Newell P., Norden C.W.: "Value of needle aspiration in bacteriologic diagnosis of cellulitis in adults". *J. Clin. Microbiol.,* 1988, *26,* 401.

[9] Mallon E., Powell S., Mortimer P., Ryan T.J.: "Evidence for altered cell-mediated immunity in postmastectomy lymphoedema". *Br. J. Dermatol.,* 1997, *137,* 928.

[10] Foeldi M.: "Treatment of lymphedema". *Lymphology,* 1994, *27,* 1.

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