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REPEATED INTRANODAL LYMPHANGIOGRAPHY FOR THE TREATMENT OF LYMPHATIC LEAKAGE

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ABSTRACT

Treatment of patients with chylous or non-chylous lymphatic leakage can be difficult. An approach using therapeutic lymphangiography can reduce the lymphatic leakage, but it seldom stops the leakage immediately and subsequent conservative treatment is necessary. We report three cases in which intranodal lymphangiography was performed multiple times to inhibit lymphatic leakage. In each case, the lymph node was punctured under ultrasound guidance using a 23-gauge needle and lipiodol was injected manually at a rate of 1 ml/3 min. The procedure was repeated twice in two cases of gastrointestinal carcinoma and four times in one case of lymphoma. In all three cases, the postoperative lymphatic leakage stopped after the repeated intranodal lymphangiography.

Keywords: lymphatic, leakage, intranodal lymphangiography, lipiodol, embolization

One of the methods utilized for the treatment of lymphatic leakage in patients is therapeutic lymphangiography (1-6). However, therapeutic lymphangiography frequently only reduces the lymphatic leakage without immediate stoppage and subsequent conservative treatment becomes necessary. We report 3 cases in which repeated intranodal lymphangiography was performed to stop lymphatic leakage and remove the need for further conservative therapy.

CASE REPORTS

Intranodal Lymphangiography

The procedure for intranodal lymphangiography was as follows. The lymph node was punctured under ultrasound guidance using a high-frequency (13-MHz) superficial linear transducer and a diagnostic ultrasound device (Prosound 3500SX and UST-5413, Hitachi Aloka Medical, Ltd., Tokyo, Japan) and a 60-mm-long, 23-gauge Cathelin needle (Terumo Europe, Leuven, Belgium). The tip of the needle was instilled at the junction between the cortex and the hilum and lipiodol was injected manually at a rate of 1 ml/3 min.

Case 1

A 61-year-old man was undergoing treatment with chemoradiation for esophageal cancer. Subsequently, esophagectomy was planned with a cervical transverse incision. The tumor was considered inoperable due to cancer invasion and the operation stopped. Two days postoperatively, a lymphocele appeared subcutaneous to the cervical incision wound causing compression symptoms which were treated with needle aspiration on an intermittent basis. Transabdominal percutaneous thoracic duct embolization was performed 21 days postoperatively, and the compression symptoms were alleviated. Although needle aspiration became unnecessary, a lymphocele dissection was

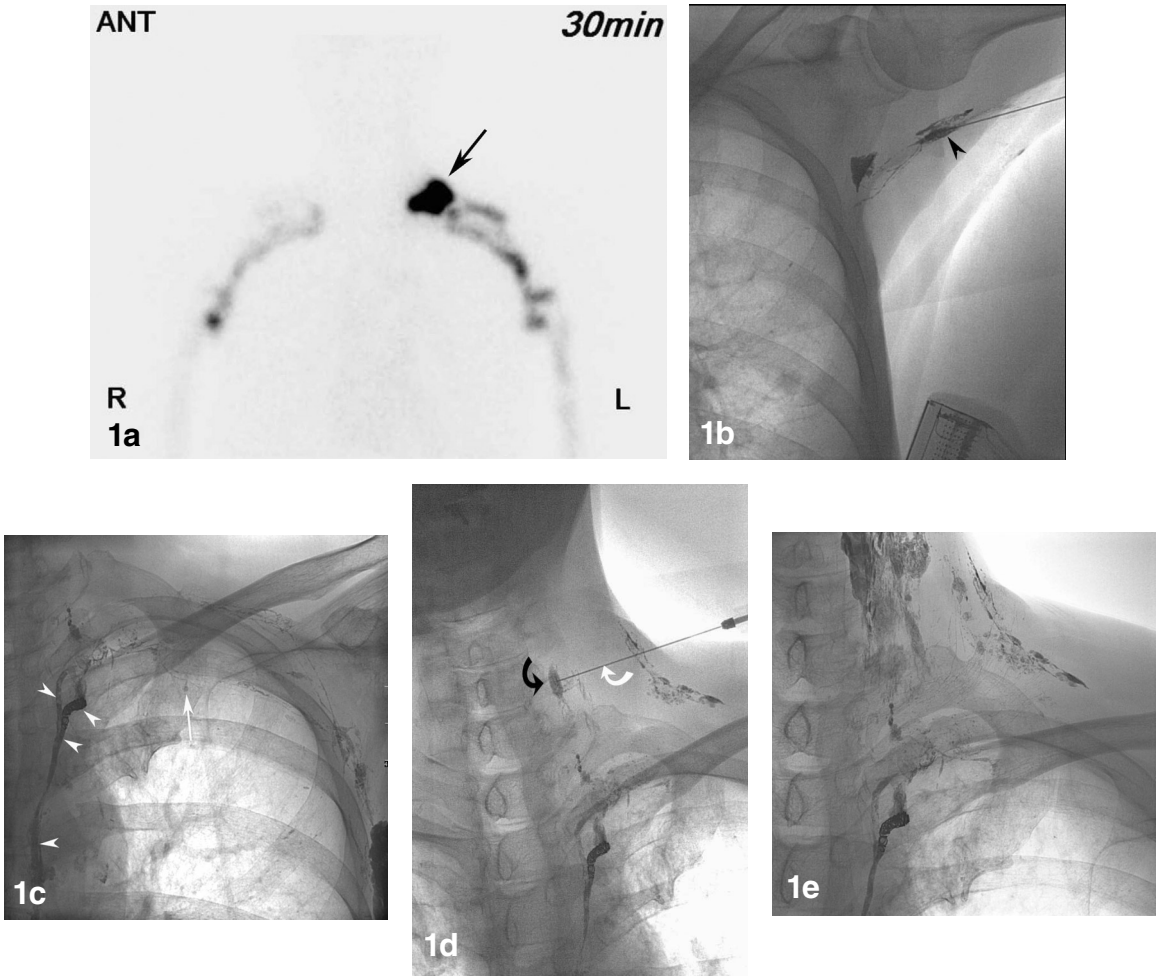

Fig. 1. A 61-year-old man underwent thoracic duct embolization to treat a lymphocele that developed after a cervical transverse incision. (a) Upper limb lymphoscintigraphy was performed with technetium-99m-labeled human serum albumin and the anterior image taken 30 minutes after injection shows leaked tracer accumulation at the left clavicle (black arrow). (b,c) The first therapeutic lymphangiography performed with lipiodol injected into a left axillary lymph node (b, black arrow head) highlighted lipiodol extravasation into the lymphocele (c, white arrow). The prior thoracic duct embolization using a coil and sclerosing agent N-butyl-2-cyanoacrylate is also seen in this image (c, white arrow heads). (d,e) Twenty-two days after the first procedure, a second therapeutic lymphangiography was performed using a left cervical lymph node (d, curved black arrow) and puncture needle (d, curved white arrow). Although lipiodol extravasation is not observed (e), lymphatic leakage stopped 2 days later.

performed because infection subsequently occurred in the residual accumulated fluid. After the infection dissipated, lymphatic leakage of ~400 ml/day continued for 75 days. Subsequently, an upper limb lymphoscintigraphy was performed with technetium-99m-labeled human serum albumin. Tracer leakage was observed extending to the lymphocele on the left side (*Fig. 1a*). Based on this finding, the first intranodal lymphangiography was performed with 5 ml of lipiodol injected by puncturing a left axillary lymph node. Lipiodol extravasation was observed from the lymphatic vessel to the central site of leakage (*Fig. 1b*). Leakage decreased to 20 ml/day after lymphan-

giography. However, due to continued leakage, a second intranodal lymphangiography was performed 22 days after the first with injection of 2 ml of lipiodol into a left cervical lymph node. Although lipiodol extravasation was not observed, lymphatic leakage from the surgical wound stopped two days later (*Fig. 1c*). There were no subsequent recurrences of lymphatic leakage.

Case 2

A 63-year-old woman underwent surgical resection and chemoradiotherapy for rectal cancer and subsequently, she underwent an ileocecal resection to treat a small intestinal perforation with a peritoneal drainage tube placement. Imaging of the drainage tube indicated a leakage of intestinal fluid through an anastomotic leak. The outflow of 400 ml/day was consistent, and after meals this fluid turned chylous indicating a component of lymphatic leak was present. Following failure of conservative treatment for 6 months, an intranodal lymphangiography was performed.

Both left and right inguinal lymph nodes were punctured, and 6 ml and 1.5 ml of lipiodol were injected in the right and left sides, respectively. Lipiodol leakage from the lymph vessel extending from the right inguinal lymph node into the drainage tube was visualized (*Fig. 2*). Following this lymphangiography, the drainage decreased to 200 ml/day. Four days later, a second intranodal lymphangiography was performed with puncture of a right inguinal lymph node with 6 ml lipiodol injected. Lipiodol extravasation was not observed, and visualization of lymph vessels joining the leakage area also decreased compared to the first lymphangiography. Drainage reduced to 100 ml/day at two days and to 40 ml/day at fourteen and drainage fluid was not chylous even postprandially. Lower limb lymphoscintigraphy was performed to verify no tracer leakage and lymphatic leakage did not recur thereafter.

Case 3

A 75-year-old man with malignant lymphoma underwent right inguinal lymphadenectomy and chemotherapy. Lymphatic leakage occurred from the surgical wound immediately postoperatively, and a Penrose drain was placed. Leakage of 300-400 mL/day was observed after the completion of chemotherapy, and the first intranodal lymphangiography was performed 60 days postoperatively utilizing a right inguinal lymph node. The second, third, and fourth intranodal lymphangiography procedures were performed 4, 10, and 17 days following the first, and lipiodol extravasation to the surgical wound was observed after each procedure (*Fig. 3*). The injected volumes of lipiodol at the first, second, third, and fourth procedures were 1.0 ml, 2.0 ml, 1.5 ml, and 1.5 ml, respectively, and the volume of lymphatic leakage was 142 ml/day, 171 ml/day, 95 ml/day, and 20 ml/day, respectively. Leakage stopped two days after the fourth lymphangiography and did not recur.

DISCUSSION

Although lymphangiography using lipiodol is successful in stopping lymphatic vessel leaks, the exact mechanism(s) of action are largely unknown. Two commonly proposed ideas are either 1) an inflammatory reaction occurring around the extravasated lipiodol causing lymphatic vessel obstruction, or 2) lipiodol plays a role as a therapeutic embolic agent inside the lymphatic vessel (1,2). Matsumoto, et al reported that lymphatic leakage stopped in 89% of patients who continued conservative treatment after lymphangiography and that the mean duration of this leakage was 17 days (range, 4-31 days) (2). Alejandre-Lafont, et al reported that cessation of lymphatic leakage was observed in 51% of patients within two weeks and in 44% of patients within one week post-lymphangiography, and a $\geq 50\%$ regression, but not cessation, in leakage was

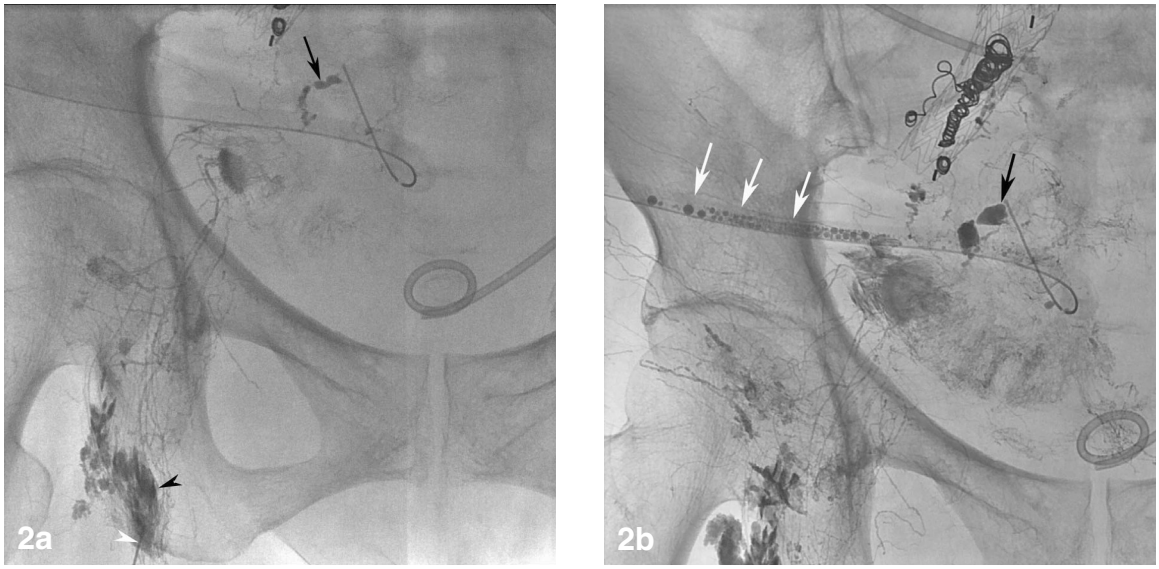

Fig. 2. A 63-year-old woman underwent ileocecal resection for a small intestinal perforation with peritoneal drainage tube placement. (a,b) Image from this first therapeutic lymphangiography with lipiodol following puncture of an inguinal lymph node (a, black arrow head) with injection needle (a and b, white arrow head) demonstrates lipiodol leaking into the pelvis (a and b, black arrow) and lipiodol within the drainage tube (b, white arrows).

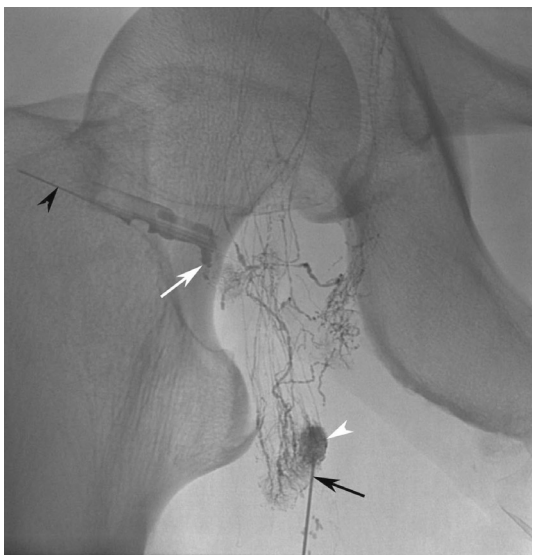

Fig. 3. 75-year-old man with malignant lymphoma underwent right inguinal lymphadenectomy resulting in lymphatic leakage of 300-400 ml/day from the surgical wound. A therapeutic lymphangiography using lipiodol and puncture of a right inguinal node was performed. The image highlights the injected lymph node (white arrow head), puncture needle (black arrow), extravasated lipiodol into the tissues (white arrow), and Penrose drain (black arrow head).

observed in 12% of the patients (3). Kos, et al reported that lymphatic leakage in 55% of the patients who underwent lymphangiography stopped after conservative treatment (4). These findings indicate that continuation of conservative treatment is necessary after therapeutic lymphangiography, and even then, the lymphatic leakage may decrease and not fully stop. Therefore, based on the mechanisms of therapeutic lymphangiography, we hypothesized that repeatedly performing lymphangiography may be effective. Previous reports have indicated the efficacy of therapeutic lymphangiography but have not recommended repeating the procedure. We postulated that one of the reasons for this may be that most reports on therapeutic lymphangiography involved bipedal lymphangiography, and repeating this procedure is complicated. A big advance came in 2012 when a report surfaced on performing intranodal lymphangiography in lieu of bipedal lymphangiography (7). Compared to bipedal lymphangiography, intranodal lymphangiography is easier, has

less morbidity, and does not require a slow infusion pump (7,8). For these reasons, we chose to perform intranodal lymphangiography to treat lymphatic leakage and to try this technique more than once in patients with continuing leakage. Our case series demonstrates three cases of postoperative lymphatic leakage that were successfully treated by performing repeated intranodal lymphangiography and this technique should be considered for further use and investigation.

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