

Single-Session Intranodal Glue Embolization for Postsurgical Refractory Groin Lymphorrhea: A Case Report

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Abstract

A 90-year-old female presented with poor right groin wound healing due to lymphorrhea and infection following a surgical cutdown procedure for arterial revascularization. Although negative pressure wound therapy (NPWT) and inguinal lymphadenectomy were performed, infection and lymphorrhea did not heal. Lymphangiography via a right inguinal lymph node revealed lymphatic leakage in the wound. Intranodal glue embolization (IGE) was performed by injecting 0.6 mL of 33% n-butyl-2 cyanoacrylate (NBCA)-lipiodol mixture. Additionally, the presence of glue in an open wound was directly confirmed in this case. After embolization, lymphorrhea ceased, and the wound healed completely. No lymphorrhea recurrence or complications were observed for 6 months. This case suggests that IGE could be an effective treatment for groin lymphorrhea.

Key words: groin lymphorrhea, lymphangiography, embolization, intranodal glue embolization

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Introduction

Inguinal lymphorrhea after arterial revascularization is a rare complication. Its onset rate is reported to be between 2% and 15% [1-3]. Prolonged hospitalization and delayed wound infection can occur when a lymphatic fistula persists in the groin area [3]. It has been reported that 70% of patients can be cured from groin lymphorrhea with conservative treatment, such as bed rest with elevation of the leg to reduce lymph flow, intravenous antibiotics, and local wound care [2]. If healing is not achieved conservatively, surgical management or negative pressure wound therapy (NPWT) is generally performed [3].

Several studies have reported that lymphangiography, using iodized oil (Lipiodol; Guerbet, Aulnay-Sous-Bois, France), could not only help diagnose but also treat lymphorrhea [4, 5].

Furthermore, recently, a successful treatment has been described by injecting a mixture of n-butyl-2 cyanoacrylate (NBCA; Histoacryl Blue; B. Braun, Melsungen, Germany) and lipiodol (glue) via an inguinal lymph node for inguinal lymphorrhea (intranodal glue embolization [IGE]) [6].

Even cases of intractable lymphorrhea can be wholly cured with single-session IGE, as in this case.

Case Report

A 90-year-old female underwent percutaneous transfemoral transcatheter aortic valve implantation (TAVI) due to severe aortic valve stenosis. The right external iliac artery accidentally was obstructed during TAVI. Therefore, revascularization under a cutdown procedure of the right groin was promptly performed. The patient had a history of Sjogren's

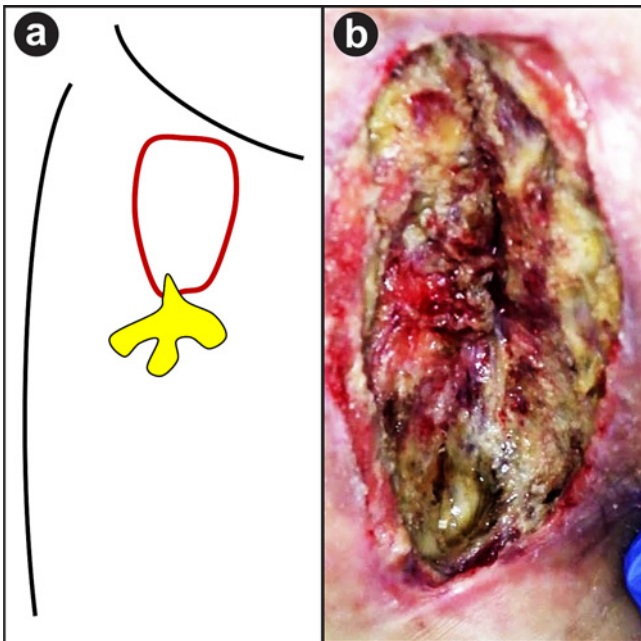


Figure 1. a: Diagram of the right groin lymphorrhhea (yellow pool) in the open wound (red circle). b: Open wound.

syndrome and rheumatoid arthritis and had been administered medications including 100 mg/day bucillamine and 5 mg/day prednisolone.

Pain and lymphorrhhea from the inguinal wound appeared after surgery. Based on the patient's clinical course after the cutdown procedure and serous fluid drainage, a diagnosis of lymphatic fistula was established. No laboratory analysis of drainage fluid was performed in this case. Moreover, lymphocele formation and cellulitis were detected by computed tomography (not shown) on the 14th day after TAVI in the right inguinal area. Therefore, for treating lymphocele, an inguinal lymph node resection surgery with ablation was performed on the 23rd day after TAVI. However, the volume of lymphorrhhea increased up to about 100-200 mL/day after reoperation. In addition, gram-negative bacilli were detected in the exudate culture, leading to a diagnosis of lymphorrhhea with infection. A 2nd reoperation for wound debridement, including lymphadenectomy of the lymph node identified as the cause of lymphorrhhea, was performed on the 33rd day after TAVI, and NPWT was initiated for the open wound.

Since infection and lymphorrhhea persisted (**Fig. 1**), the cardiovascular surgeons asked for lymphangiography. The patient provided informed consent for the lymphangiography and embolization procedures. An institutional review board approval was obtained for the use of NBCA.

On the 38th day after TAVI, ultrasound-guided intranodal lymphangiography with lipiodol was performed using 10 MHz linear probe, 25 G 60-mm needle, and extension tube (50 cm length, 1.0 mm inner diameter, 0.4 mL lumen) (**Fig. 2**). This procedure was performed via a right inguinal lymph node, which was located slightly caudal from the open wound. It demonstrated leakage of lipiodol in the open wound (**Fig. 3**). Thus, the decision was made to perform

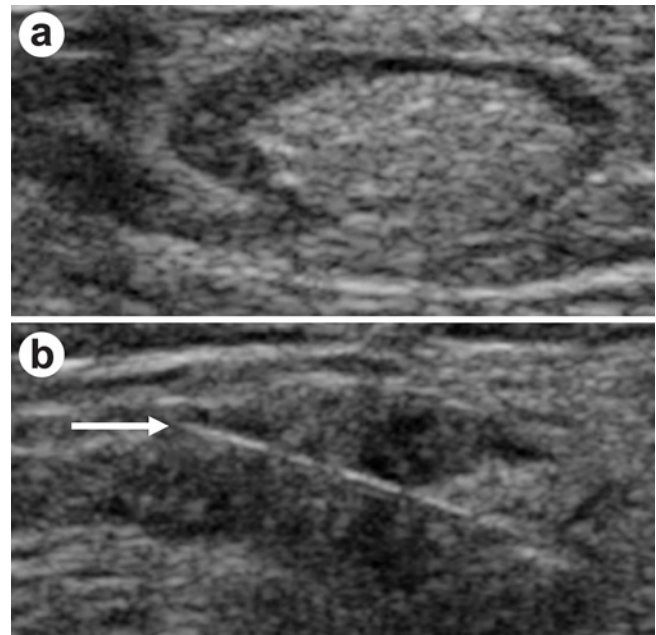


Figure 2. Ultrasound (10 MHz linear probe) images. a: Right inguinal lymph node caudal to the open wound. b: Puncturing of the lymph node using 25 G 60-mm needle (white arrow).

IGE. Lipiodol was washed out by injecting a liquid containing 5% glucose and 1% lidocaine for pain relief purposes in the ratio of 9:1. Thereafter, 0.6 mL of 33% NBCA-lipiodol mixture (glue) was slowly administered under fluoroscopy until glue sufficiently filled the efferent lymphatic vessels up to the leakage point (**Fig. 3**). After IGE, the presence of glue in the open groin wound was demonstrated visually (**Fig. 4**). The total procedure time was half an hour.

Lymphorrhhea disappeared completely a day after this IGE. Thereafter, NPWT was continued to cure infection and the wound. The wound healed, and the patient was discharged 95 days post-TAVI. No lymphorrhhea recurrence or complications were observed for 6 months after IGE.

Discussion

Surgical ligation of lymphatic vessels is an effective option for lymphorrhhea. However, it has potential technical difficulties in identifying damaged lymphatic vessels [7]. In this case, surgical lymphadenectomy and ablation were performed in addition to NPWT. However, they were ineffective. It was suggested that combination treatments failed in this case due to prolonged wound infection and healing after the administration of steroids. In addition, this failure resulted from intractable lymphorrhhea due to the weak tissue pressure of the wound caused by the patient's advanced age.

Intranodal lymphangiography using lipiodol via an inguinal lymph node plays a role in identifying lymphorrhhea in about 90% of cases [8]. Lymphorrhhea is sometimes cured by lymphangiography alone due to its ability to cause mild inflammation [8]. However, the treatment effect of lymphangi-

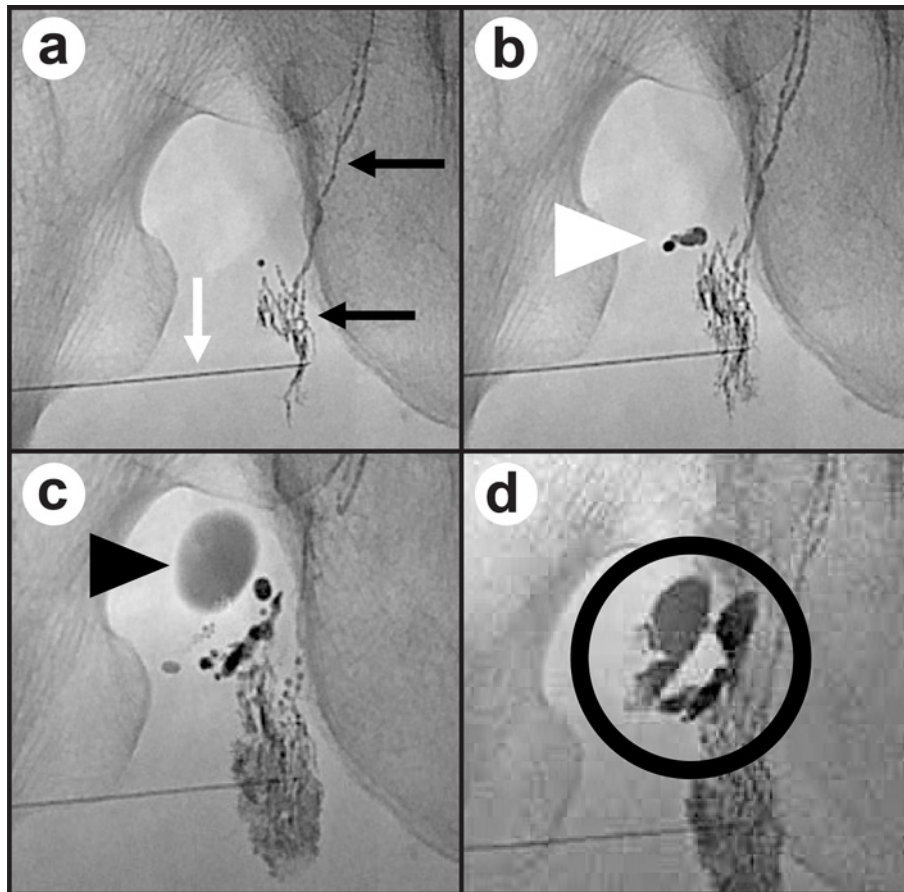


Figure 3. Fluoroscopic images of lymphangiography and intranodal glue embolization. **a:** Puncturing of the right inguinal lymph node caudal to the open wound using 25 G 60-mm needle (white arrow); lipiodol was then injected (black arrow). **b:** Accumulation of lipiodol in lymphorrhea from lymphatic duct (white arrow head). **c:** Lymphangiography demonstrated lipiodol leakage (black arrow head) in the open wound. **d:** Intranodal glue embolization resulted in the accumulation of NBCA–lipiodol mixture at the site of lymphorrhea in the open wound (black circle).

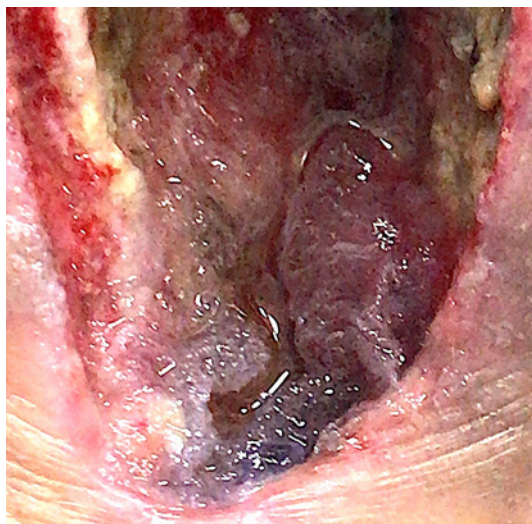


Figure 4. Open wound showing bluish-violet discoloration due to NBCA accumulation.

ography is not necessarily promising.

Recently, IGE with glue using intranodal lymphangiogra-

phy has been reported to embolize lymphorrhea [6, 9]. In the present case, single-session IGE could obstruct the intractable lymphorrhea within a shorter treatment period (24 hours after treatment). Chick et al. reported a successful case of IGE for inguinal lymphorrhea, which ceased completely after single-session IGE [9]. On the other hand, another report demonstrated a clinical success rate of 80% for IGE in 10 cases of inguinal lymphorrhea and lymphocele [6]. Only 20% (2 of 10) of these patients were healed 24 hours after IGE. In this report of 10 patients, the median time to resolution was 7 days (range, 1-17) after IGE [6]. In cases that take long time to heal, there may be insufficient accumulation of NBCA in disrupted lymphatic vessels that caused lymphorrhea, and lymphatic flow could be only decreased, not disappear completely. This delayed healing might also be due to the fact that a small amount of lymphorrhea from the collateral lymph vessels could have remained after embolization of the main lymphatic vessels by intralymphatic pressure in the upstream vessels.

The inguinal nodes range from 10 to 20 in number and are distributed widely in the groin area [3]. Therefore, it seems that many lymph nodes and efferent lymphatic ducts

might have been involved in the formation of groin lymphorrhea. In fact, in 40% of cases, IGE for groin lymphorrhea was required two to four times [6].

In the present case, lymphorrhea healed entirely with a single-session IGE. This might be explained by that most of groin lymphorrhea-related lymph nodes had been already removed by surgical resection before IGE. Furthermore, the glue leaked into the open wound, confirming that it sufficiently filled the efferent lymphatic vessels until the leakage point. As a result, lymphorrhea healed soon after a single-session IGE in this case.

Gently injecting a 33%-50% NBCA-lipiodol mixture was important for successful IGE [6, 10], and the dose of glue mixture has been reported to be about 1 mL per session of IGE in cases of groin lymphorrhea and lymphocele [6]. To successfully inject glue to fill the leakage point of the lymphatic duct that was damaged, it seems that the appropriate puncture point might represent the border between the cortex and the medulla of the lymph node. A precise puncturing of the lymph node with ultrasound is essential to ensure that the puncture needle does not deviate from the lymph node due to pressure at the time of glue injection. Generally, precise lymphangiography image interpretation is helpful to confirm the disrupted part of the downstream lymphatic ducts of the punctured lymph node. Also, to fill the embolic material adequately, flushing out the lipiodol stored at the disrupted part before embolization is important. Therefore, we performed embolization with glue after washing out lipiodol using 5% glucose that was X-ray transmissive [10].

In conclusion, as in this case, IGE could be an effective treatment for refractory groin lymphorrhea.

Conflict of Interest: None

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Informed Consent: Written informed consent was obtained from

the patient for the publication of this case report and any accompanying images.

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