

# Venous insufficiency, lymphocutaneous fistula, and use of autologous blood

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## ABSTRACT

Lymphocutaneous fistulas are an uncommon occurrence that can present after surgical intervention in lymphatic-rich areas. A spontaneous lymphatic leak can occur in conjunction with lymphedema or in the vicinity of venous stasis ulcers. However, spontaneous lymphocutaneous fistulas are extremely rare. Traditional treatments have included surgical ligation, negative pressure therapy, and embolization. We present the case of a lymphocutaneous fistula secondary to chronic venous stasis, which was treated via injection of autologous blood and subsequent thrombosis. In the present case report, we have demonstrated successful embolization of a lymphatic vessel via autologous blood injection, a nontraditional technique that, nonetheless, yielded positive and lasting results. (*J Vasc Surg Cases and Innovative Techniques* 2021;7:61-3.)

**Keywords:** Autologous blood; Lymphatic leak; Lymphedema; Lymphocutaneous; Stasis; Venous insufficiency

## CASE REPORT

A 95-year-old woman with a medical history significant for chronic venous insufficiency had presented to the vascular surgery clinic with a 1-year history of a nonhealing wound on the medial aspect of the right ankle and persistent clear drainage from a single small opening in the skin adjacent to the wound. Drainage increased in the dependent position and had increased to  $\leq 500$  mL/d, although it could be halted by application of gentle pressure. The patient had been using thigh-length compression stockings at 20 to 30 mm Hg for several months without improvement in her symptoms. On examination, she had palpable distal pulses and normal ankle brachial indexes bilaterally. On further examination of the wound, a single lymphatic vessel was identified, which had eroded to form a fistula tract to the skin (Fig 1). A venous ultrasound scan of the lower extremity was positive for reflux in the great saphenous vein (GSV) with a reflux time of 2215 ms. The patient provided written informed consent for publication of her case details and images.

The patient underwent endovenous laser therapy of the GSV in the clinic setting, and a postoperative ultrasound scan showed successful occlusion of the GSV. On removal of compression, drainage from the lymphocutaneous fistula resumed. To map

the course of the lymphatic vessel, diluted povidone-iodine was gently injected into the fistula and was observed to fill the course of the vessel. After 15 minutes, the drainage had not stopped. Thus, 1% polidocanol foam was then injected into the vessel (Fig 2). Manual compression was continued for 5 minutes after injection; however, the drainage had continued on release of pressure. At 15 minutes after this second failed attempt at occlusion, 2 mL of the patient's blood were drawn from the cephalic vein and injected through the drainage site until blood was observed to fill the entire visible course of the lymphatic vessel (Fig 3). After 5 minutes of manual compression, the drainage had stopped (Fig 4). The patient was discharged from the clinic with 20- to 30-mm Hg compression stockings with a simple noncompressive adherent dressing over the fistula site. She presented for follow-up examination 2 weeks after procedure, and her stasis ulcer had healed. No further lymphatic drainage was noted at the follow-up examinations at both 2 weeks and 3 months.

## DISCUSSION

Lymphoceles and lymphocutaneous fistulas are rare, but well-described, complications that have been most often reported after vascular surgery in the pelvis or inguinal region.<sup>1,2</sup> Various techniques have been used in the treatment of postoperative lymphatic complications, including surgical exploration and drainage, sclerosing agents such as doxycycline, polidocanol, or povidone-iodine, and vacuum-assisted closure therapy.<sup>2-6</sup> Although a wide range of data have been reported on the management of postoperative lymphatic complications, very little has been reported on the management of such fistulas that occur outside the postoperative setting. Our patient did not have a history of lower limb surgery. However, her history of chronic venous insufficiency likely contributed to the increased pressure in the lymphatic vessels of the lower extremity and subsequent lymphocutaneous fistula formation.

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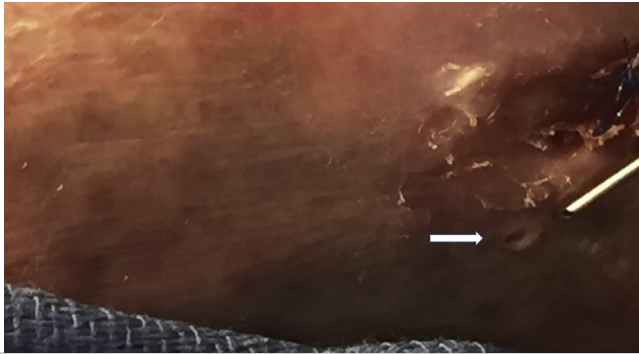
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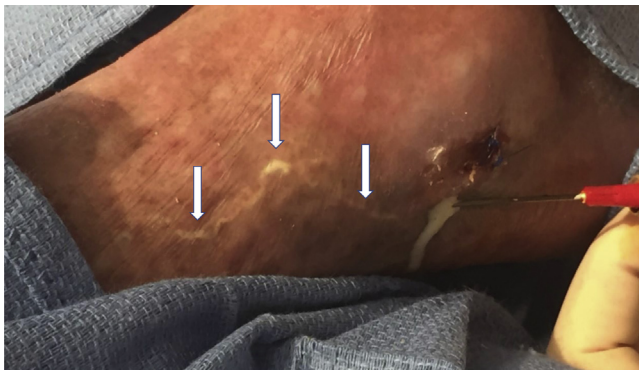
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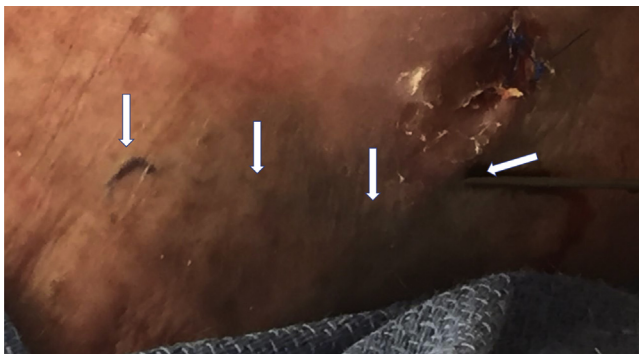
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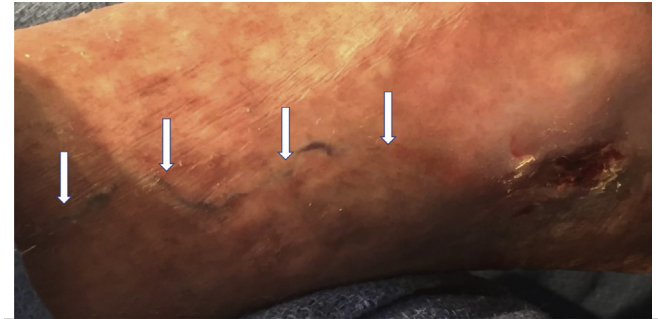
**Fig 1.** Opening of the lymphatic vessel (arrow).



**Fig 2.** Injection of polidocanol (arrows).



**Fig 3.** Injection of autologous blood (arrows).



**Fig 4.** Thrombosis and cessation of the lymphatic leak (arrows).

An epidural blood patch is a well-described treatment for headaches resulting from cerebrospinal fluid leaks after lumbar puncture.<sup>7</sup> During this procedure, a small volume of autologous blood is injected into the epidural space to stop the leak of cerebrospinal fluid.<sup>8</sup> By extrapolation of this technique to our clinical scenario, we aimed to use the patient's blood in a similar fashion to embolize her lymphatic fistula. We discussed with the patient that this technique had not been previously used in this setting. However, the risk of adverse effects from the patient's own blood were minimal. The patient agreed to attempt this technique after a discussion of the risks and potential benefits. The choice of an autologous blood injection was considered a less-invasive option that could be performed in the clinic setting with minimal extra preparation. By using the patient's own blood, we were able to embolize the lymphatic vessel without a prolonged course of drainage and the risk of future reintervention. We postulated that this novel method worked via a twofold mechanism—the inflammation and endothelial damage to the lymphatic vessel from the povidone-iodine and polidocanol facilitated easier clot formation when the patient's blood was injected. This allowed for sclerosis and permanent closure from subsequent fibrosis after coagulation. Agents such as bleomycin and doxycycline have been described in the closure of congenital lymphatic malformations with a similar mechanism involving nonspecific inflammation resulting in eventual sclerosis. However, these require multiple injections spaced apart over a period of weeks.<sup>9</sup> To the best of our knowledge, the present report is the first to describe a lymphocutaneous fistula embolization via autologous blood injection.

## CONCLUSIONS

We have reported a unique case of a spontaneous lymphocutaneous fistula that was embolized via injection of autologous blood. This represents a nontraditional technique that was attempted after other accepted techniques for fistula closure had failed. Lymphocutaneous fistulas that present spontaneously without an associated surgical history are extremely rare, but nonetheless

Our patient's lymphatic drainage persisted even after multiple attempts to occlude the lymphatic vessel, first with povidone-iodine and, subsequently, with 1% polidocanol. Given our patient's history of venous insufficiency and the proximity of the lymphatic opening to the non-healing stasis ulcer, more invasive treatment options such as ligation of the lymphatic channel and surgical incision and drainage were deemed unsuitable owing to the risk of impaired wound healing. Likewise, she was unsuitable for vacuum-assisted closure therapy owing to the lack of an open wound that could be packed with a sponge.

can be managed by similar techniques to fistulas that present postoperatively.

## REFERENCES

1. Schwartz MA, Schanzer H, Skladany M, Haimov M, Stein J. A comparison of conservative therapy and early selective ligation in the treatment of lymphatic complications following vascular procedures. *Am J Surg* 1995;170:206-8.
2. Tyndall SH, Shepard AD, Wilczewski JM, Reddy DJ, Elliott JP, Ernst CB. Groin lymphatic complications after arterial reconstruction. *J Vasc Surg* 1994;19:858-64.
3. Caliendo MV, Lee DE, Queiroz R, Waldman DL. Sclerotherapy with use of doxycycline after percutaneous drainage of postoperative lymphoceles. *J Vasc Interv Radiol* 2001;12:73-7.
4. Hackert T, Werner J, Loos M, Büchler MW, Weitz J. Successful doxycycline treatment of lymphatic fistulas: report of five cases and review of the literature. *Langenbecks Arch Surg* 2006;391:435-8.
5. Klode J, Klötgen K, Körber A, Schadendorf D, Dissemmond J. Polidocanol foam sclerotherapy is a new and effective treatment for post-operative lymphorrhea and lymphocele. *J Eur Acad Dermatol Venereol* 2010;24:904-9.
6. Van den Brande P, von Kemp K, Aerden D, Debing E, Vanhulle A, Staelens I, et al. Treatment of lymphocutaneous fistulas after vascular procedures of the lower limb: accurate wound reclosure and 3 weeks of consistent and continuing drainage. *Ann Vasc Surg* 2012;26:833-8.
7. Gaiser RR. Postdural puncture headache: an evidence-based approach. *Anesthesiol Clin* 2017;35:157-67.
8. Tubben RE, Jain S, Murphy PB. Epidural blood patch. In: StatPearls [Internet]. Treasure Island, Fla: StatPearls Publishing; 2020. Available at: <http://www.ncbi.nlm.nih.gov/books/NBK482336/>. Accessed April 28, 2020.
9. Legiehn GM, Heran MKS. A step-by-step practical approach to imaging diagnosis and interventional radiologic therapy in vascular malformations. *Semin Intervent Radiol* 2010;27:209-31.

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