# Treatment of Lymphorrhea Associated with an Amputation Stump with Lymphaticovenular Anastomosis

# Tratamento de Linforreia Associada a um Coto de Amputação com Anastomoses Linfatico-Venosas



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#### **ABSTRACT**

Lower limb lymphorrhea is a condition with a considerable impact on the quality of life. It is usually associated with inguinal lymph node dissection and vascular procedures with femoral exposure. In this case report, we describe a patient who underwent a below-knee amputation and two years later developed lymphorrhea from the stump, preventing adaptation to the prosthesis. Lymphoscintigraphy showed a delayed lymphatic progression. After failure of conservative treatment, she underwent lymphaticovenular anastomosis with a successful outcome. Drainage cessation suggests that lymphaticovenular anastomosis may be an effective treatment for patients with lymphorrhea from and amputation stump, although further studies are required to determine long-term efficacy.

Keywords: Amputation Stumps; Anastomosis, Surgical; Lower Extremity; Lymphatic Vessels; Microsurgery

## **RESUMO**

A linforreia do membro inferior é uma patologia com grande impacto na qualidade de vida. Está geralmente associada a esvaziamentos ganglionares inguinais e a procedimentos vasculares com exposição dos vasos femorais. Apresentamos o caso de uma doente que sofreu uma amputação abaixo do joelho e dois anos depois desenvolveu linforreia a partir do coto de amputação, impedindo a adaptação à prótese. A linfocintigrafia revelou um atraso na progressão linfática. Após falência do tratamento conservador, foi submetida a anastomoses linfático-venosas, com resolução da linforreia. A cessação da drenagem sugere que a realização de anastomoses linfático-venosas poderá ser um tratamento eficaz em doentes com linforreia a partir de um coto de amputação, embora sejam necessários mais estudos para determinar a sua eficácia a longo prazo.

Palavras-chave: Anastomose Cirúrgica; Cotos de Amputação; Extremidade Inferior; Microcirurgia; Vasos Linfáticos

#### INTRODUCTION

Lower limb lymphorrhea and lymphoceles are known complications of any trauma or procedure that disrupts the lymphatic system in the inguinal or femoral region. Lymphaticovenular anastomosis are an effective surgical option in these early onset complications. <sup>1-5</sup> We present a rare case of late onset lymphorrhea and generalized lymphatic system impairment after a below-knee amputation, treated with lymphaticovenular anastomosis.

# **CASE REPORT**

The patient was a 30-year-old woman who underwent a below-knee amputation due to a high energy trauma of the leg with extensive soft tissue damage, comminution and arterial damage (Gustilo IIIC) after suffering a road accident at age 26 (Fig. 1). She also suffered a fracture of the ipsilateral femur treated with an external fixator. The stump was skin grafted due to severe soft tissue trauma of the leg and one month later complete healing was achieved. Lymphorrhea from the stump began two years later, which prevented adaptation to the prosthesis and was associated with an episode of lymphangitis. On examination she presented several invaginated and macerated areas on the skin grafted stump where the lymph was coming from. Lymphoscintigraphy showed a delayed lymphatic progression, characterized by lower impregnation intensity of the ipsilateral lymph nodes. After 11 months of conservative therapy with manual lymphatic drainage and customized compression garment with no improvement, lymphaticovenular anastomosis was proposed to the patient. After performing indocyanine green lymphangiography (we diluted 25 mg indocyanine green with 2.5 mL of sterile water and injected in three different sites approximately 0.3 mL for a total of 1 mL), two channels that showed splash pattern of dermal backflow were selected, one in the medial thigh over the ventromedial bundle (Fig. 2) and the other laterally, over a local collector. Patent blue was injected 5 cm distally at each location. Two end-to-end lymphaticovenular anastomosis were performed with 10 - 0 and 11 - 0 nylon through 2 cm incisions, on both the medial and lateral aspects of the thigh (Fig. 3). Two weeks after surgery the patient resumed conservative treatment and knee flexion was encouraged. In the postoperative period the patient had multiple blisters in the stump and a slight enlargement of the limb. Two months later, the blisters healed and no more drainage was detected. The use of the prosthesis was delayed up to four months postoperatively due to hypersensitivity in the scar of the lymphaticovenular anastomosis on the medial aspect of the thigh. At 24 months follow-up the patient remained adapted to the prosthesis without any trace of drainage.

#### **DISCUSSION**

This patient presented an impaired lymph transport with

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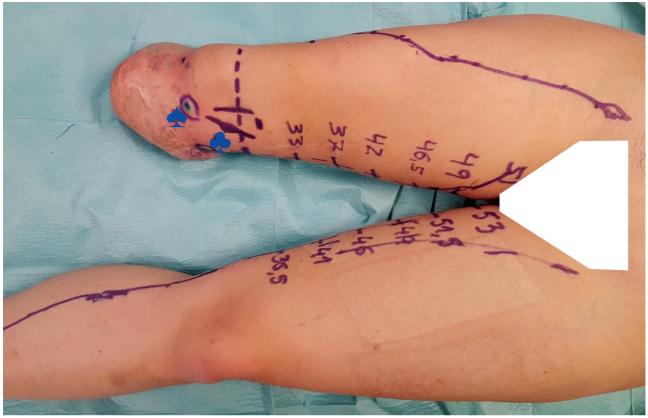


Figure 1 – Record of the perimeter of the limbs every 4 cm (♣ indicates the location of the lymphaticovenular anastomosis on the medial aspect of the thigh above the knee; ♣ indicates the point of patent blue injection).

no edema – (stage 0) according to the International Society of Lymphology. No swelling was present due to continued discharge of lymph. In fact, the right thigh had a lower volume than compared to the left thigh, probably due to muscular atrophy due to inactivity.

Microsurgical reconstructive techniques for lymphede-

ma are limited to the early stages of the disease. When the fluid dominant edema progresses to adipose dominant edema and then to fibrosclerotic dominant edema, surgical options are limited to liposuction and direct excision respectively. Lymphaticovenular anastomosis can be performed as long as there are functioning lymphatics.<sup>6</sup> This



Figure 2 – Splash pattern on the medial aspect of the thigh above the knee

procedure has also been shown to have positive results in cases of inguinal or femoral lymphorrhea and lymphoceles after disruption of the lymphatic system, such as inguinal lymph node biopsy or dissection, vascular procedures with exposure of femoral vessels, harvesting of superficial circumflex iliac artery perforator flap and resection of sarcomas of the adductor compartment. 1-3,5 These cases represent a localized iatrogenic blockage of lymphatic drainage, leaving distal lymphatics open. This can occur also after trauma, which could explain the occurrence of chronic wounds with persistent drainage.4 Our case presents a different mechanism, since there was a generalized delayed lymphatic progression, without a localized blockage, and the onset of lymphorrhea was not immediately after amputation. The etiology is also different from the cases previously mentioned, as there were no open distal lymphatics feeding the drainage. As far as we know, no article has been published regarding the applicability of lymphaticovenular anastomosis in the treatment of lymphorrhea from an amputation stump.

We preferentially perform lymphaticovenular anastomosis on the medial aspect of the thigh immediately above the

knee (Fig. 1). This location, proposed by Yukio Seki *et al*, allows the use of large and high-flow lymphatics, deep to the superficial fascia, with diameters larger than 0.65 mm in 60% of cases. Knee joint movement works as a pump, directing lymph flow and decreasing the risk of regurgitation or occlusion of the lymphaticovenular anastomosis. Indocyanine green lymphangiography does not usually detect lymphatics in this layer of subcutaneous tissue, deeper than 2 cm from the skin surface. Nevertheless, we performed the test and we were able to visualize fluorescence. Although the patient was thin, we admitted that it was a superficial lymphatic with a good caliber, hypothesis that was confirmed during surgery.

This patient had two perpendicular scars on the medial aspect of the thigh in the distal third, which is probably a consequence of external fixator placement. Given the high probability of injury to the lymphatic system at this level, although the lymphoscintigraphy did not show a localized blockage, it was of the utmost importance to perform lymphaticovenular anastomosis distally. Fortunately, the lymphangiography revealed lymphatics at this level.



Figure 3 – Lymphaticovenular anastomosis on the medial aspect of the thigh above the knee, superficial to the superficial fascia. Ly: lymphatic; Ve: venule

The second lymphaticovenular anastomosis was made on the lateral aspect of the thigh with a local collector with a diameter of about 0.4 mm.

Anastomoses should be performed distally to the injury, thus diverting the lymphatics responsible for persistent drainage. Considering that in this case the injury was only located at the level of amputation, and not at the level of the scar, and therefore the lymphorrhea occurred due to reflux in the proximal lymphatics, this case reports a different etiology and approach. Although lymphoscintigraphy did not show a block at the level of the referred scar but instead a delayed lymphatic progression, we still chose to perform an anastomosis distally, following what has been published regarding this subject, although with a different clinical scenario.

In the postoperative period the patient had multiple blisters in the stump and a slight enlargement of the limb attributable to the cessation of drainage. The hypersensitivity in the scar may have been due to the fact that the medial lymphaticovenular anastomosis was made in the path of the saphenous nerve.

At 24 months follow-up the patient continued to adapt to the prosthesis without any trace of drainage. The patient even became pregnant and throughout pregnancy remained uneventful.

Lymphaticovenular anastomosis may be an effective treatment for patients with lymphorrhea from and amputation stump, although further studies are required to determine long-term patency and efficacy.

## **AUTHORS CONTRIBUTION**

FMC, MM, AA: Substantial contributions to the conception or the acquisition, analysis or interpretation of data for the work. Drafting the work or revising it critically for impor-

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#### PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in 2013.

#### **DATA CONFIDENTIALITY**

The authors declare having followed the protocols in use at their working center regarding patients' data publication

#### PATIENT CONSENT

Obtained.

# **COMPETING INTERESTS**

All authors report no competing interests.

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