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Case Report | Olgu Sunumu

REHABILITATION APPROACH IN THE TREATMENT OF ADVANCED CONGENITAL LYMPHEDEMA AND LYMPHORRHEA WITH IMPROVED MUSCLE OXYGENATION: A CASE REPORT

İLERİ EVRE KONJENİTAL LENFÖDEM VE LENFORE TEDAVİSİNDE KAS OKSİJENLENMESİNİ ARTIRAN REHABİLİTASYON YAKLAŞIMI: OLGU SUNUMU

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ABSTRACT

Lymphedema is a distinct form of edema characterized by its chronic and progressive course, resulting from impaired lymphatic formation and drainage. Both congenital and acquired abnormalities of the lymphatic system can lead to the accumulation of macromolecules within the interstitial space and subsequent fluid retention. Lymphorrhea accompanying congenital lymphedema is rare, and studies evaluating its effects on muscle oxygenation are limited in the literature. This case report describes the effects of a comprehensive rehabilitation program in a 52-year-old woman with advanced congenital lymphedema and lymphorrhea, emphasizing changes in muscle oxygenation. The patient, who had lifelong lower limb swelling and mobility restriction, was diagnosed with stage 2 congenital lymphedema and lymphorrhea and participated in a six-week rehabilitation program comprising manual lymphatic drainage, skin care, exercise, and multilayer compression bandaging performed five times per week. Following treatment, limb circumference decreased, joint range of motion improved, and muscle oxygenation assessed by near-infrared spectroscopy showed notable enhancement. Additionally, the patient lost 12 kg, was able to walk 1 km without pain, and reported an improved quality of life. These findings demonstrate that comprehensive rehabilitation approaches involving manual lymphatic drainage can significantly contribute to not only edema control but also improved muscle oxygenation and functional capacity in congenital lymphedema with lymphorrhea.

Keywords: lymphedema, lymphorrhea, manual lymphatic drainage, muscle oxygenation

ÖZ

Lenfödem, kronik ve ilerleyici bir yapıya sahip, lenf oluşumu ve drenajındaki bir bozukluktan kaynaklanan özel bir ödem türüdür. Lenfatik sistemdeki doğuştan veya sonradan edinilmiş bir anormallik, interstisyel alanda makromoleküllerin birikmesine ve sıvıların tutulmasına neden olur. Konjenital lenf ödemle birlikte lenfore nadir görülmekte olup, kas oksijenlenmesi üzerindeki etkilerini değerlendiren çalışmalar literatürde sınırlıdır. Bu olgu sunumu, ileri evre konjenital lenfödem ve lenfore tanılı 52 yaşındaki bir kadında uygulanan kapsamlı bir rehabilitasyon programının etkilerini ve kas oksijenlenmesindeki değişimleri incelemektedir. Alt ekstremite şişliği ve hareket kısıtlılığı bulunan hasta, haftada 5 kez uygulanan manuel lenf drenajı, cilt bakımı, egzersiz ve çok katmanlı kompresyon bandajlamasından oluşan altı haftalık bir rehabilitasyon programına katılmıştır. Tedavi sonunda, ekstremite çevresinde azalma, eklem hareket açıklığında artış ve yakın kızılötesi spektroskopi (NIRS) ile ölçülen kas oksijenlenmesinde belirgin iyileşme gözlenmiştir. Ayrıca hasta 12 kg kilo kaybı sağlamış, ağrısız şekilde 1 km yürüyebilmiş ve yaşam kalitesinde belirgin artış bildirmiştir. Bu bulgular, manuel lenf drenajını içeren kapsamlı rehabilitasyon yaklaşımlarının, lenfore ile seyreden konjenital lenfödemde yalnızca ödem kontrolü değil, aynı zamanda kas oksijenlenmesi ve fonksiyonel kapasitenin iyileştirilmesinde de önemli katkılar sağlayabileceğini göstermektedir.

Anahtar kelimeler: lenfödem, lenfore, manuel lenf drenajı, kas oksijenasyonu

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Introduction

Lymphedema is a chronic and often debilitating condition characterized by the accumulation of protein-rich interstitial fluid due to impaired lymphatic drainage, leading to progressive swelling, tissue fibrosis, and functional impairment .[1] Lymphedema is categorized as congenital primary when it presents before two years of age, as early primary between two and 35 years, and as late primary when onset occurs after 35 years of age.[2] Epidemiological data on the prevalence of lymphedema are limited; however, it is estimated that congenital primary lymphedema affects approximately 1.15 per 100,000 children in the United States. A population-based study reported the prevalence of chronic edema to be 1.33 per 1,000 individuals, rising to 5.4 per 1,000 among those aged 65 years and older.[3]

Congenital lymphedema often goes unnoticed until it has progressed, making diagnosis and management significantly more challenging.[2] A conservative approach to lymphedema treatment involves several complex physical therapy methods, including manual lymphatic drainage, skin care, exercise, and compression treatments using elastic bandages.[3] These treatments improve lymphatic circulation, facilitating the removal of metabolic waste and interstitial fluid, which increases capillary blood flow and promotes oxygen delivery to tissues. This increased oxygenation supports cellular repair, reduces inflammation, and improves overall tissue metabolism. [4] However, data on measuring the increase in muscle oxygenation following conservative treatments of lymphorrhea associated with the rare congenital lymphedema using current technologies are almost nonexistent in the literature. Nevertheless, challenges persist regarding early diagnosis, ensuring long-term adherence to treatment, and tailoring therapeutic approaches to individual needs.[3–4]

In this case report, we aim to present the effects of a physiotherapy program applied in the treatment of a patient with advanced lymphedema accompanied by lymphorrhea on edema, muscle oxygenation, and functional capacity.

Case Report

Examination and Clinical Presentation

History

A 52-year-old female patient, with a long-standing history of lower limb edema and mobility impairment dating back to childhood, has been diagnosed with stage 2 congenital lymphedema accompanied by lymphorrhea. This diagnosis highlights the progressive nature of her condition, which is characterized by an abnormal accumulation of lymphatic fluid leading to significant functional limitations and potential complications.

At presentation, she reported severe pain, restricted mobility, and leakage of lymphatic fluid. The patient was evaluated by a physician at Haymana State Hospital in Ankara. She revealed a history of symptomatic lymphedema that began in childhood around the age of 10, yet she had not received any treatment for it. In the five years preceding her intervention, she consulted medical professionals for lower extremity edema but was informed that no effective treatment options were available. Furthermore, she had not undergone any diagnostic testing for lymphedema. The patient conveyed feelings of depression and social stigma associated with the size of her legs, which hindered her from wearing “normal” clothing and shoes. She also noted a significant increase in edema in her left lower extremity approximately one year prior to her referral for lymphedema treatment.

Systems review

The patient was diagnosed with stage 2 lymphedema in March 2025. Upon examination, findings included dryness, porosity, hardness, and skin fibrosis in her legs. Her range of motion was severely restricted, and for the past year, she had been unable to walk more than 10 meters due to significant swelling and pain. Additionally, she could only bend her knees to 25°, which greatly affected her quality of life. The patient also had a notable medical history of a herniated lumbar disc. A comprehensive medical examination was conducted, and all contraindications for lymphedema treatment were ruled out before she commenced specialized physical therapy. Although the patient did not show symptoms of arterial or venous insufficiency, vascular assessments, including arterial and venous Doppler examinations, were performed initially. An appropriate physiotherapy program was developed for her, and she was referred to physiotherapy. All procedures were carried out by a specialized physiotherapist certified in lymphedema at Ankara University Haymana Vocational School. The patient was informed that data and photographs related to treatment would be submitted for publication, and consent was obtained (Figure 1).

Figure 1. The patient's legs before treatment

Tests and measures

Both lower extremities exhibited edema, resulting in a considerable limitation in range of motion. Standard circumference measurements were performed using a Jobst/Alvarex measuring board equipped with a footplate attachment. While the patient was in a supine position, the footplate was positioned against the sole, and the measuring board was placed underneath the lower extremity. Circumference measurements were recorded at the metatarsals, midfoot, and heels. Vertical measurements of the lower extremity were also taken in the supine position, specifically at the 5 cm mark, using the graduated scale on the Jobst/Alvarex board. [6] Measurements were obtained at 10 cm intervals until reaching the mid-thigh. Both lower extremities exhibited edema. The patient demonstrated moderate to severe edema in the left lower extremity, with a significant 36 cm discrepancy in measured increments compared to the right lower extremity. Joint range of motion was assessed using a goniometer (Tables 1, 2).

Table 1. Lymphedema measurements chart

İntervals	Side	Week 0	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6
17–Metatarsal	Right	27.0	25.0	23.0	21.0	19.0	17.0	15.0
	Left	29.0	27.0	25.0	23.0	21.0	19.0	17.0
12–Mid-foot	Right	32.2	30.2	28.2	26.2	24.2	22.2	20.2
	Left	32.0	30.0	28.0	26.0	24.0	22.0	20.0
Heel	Right	37.0	35.0	33.0	31.0	29.0	27.0	25.0
	Left	40.4	38.4	36.4	34.4	32.4	30.4	28.4
5 cm	Right	31.9	29.9	27.9	25.9	23.9	21.9	19.9
	Left	33.3	31.3	29.3	27.3	25.3	23.3	21.3
10 cm	Right	33.4	31.4	29.4	27.4	25.4	23.4	21.4
	Left	34.5	32.5	30.5	28.5	26.5	24.5	22.5
20 cm	Right	39.8	37.8	35.8	33.8	31.8	29.8	27.8
	Left	48.0	46.0	44.0	42.0	40.0	38.0	36.0
30 cm	Right	45.6	43.6	41.6	39.6	37.6	35.6	33.6
	Left	57.0	55.0	53.0	51.0	49.0	47.0	45.0
40 cm	Right	42.2	40.2	38.2	36.2	34.2	32.2	30.2
	Left	51.5	49.5	47.5	45.5	43.5	41.5	39.5
50 cm	Right	47.6	45.6	43.6	41.6	39.6	37.6	35.6
	Left	55.0	53.0	51.0	49.0	47.0	45.0	43.0

Table 2. Measurements of range of motion

Rom	Week 0	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6
Knee Right	30 °	45 °	90 °	100 °	120 °	130 °	140 °
Knee Left	45 °	60 °	95 °	105 °	125 °	145 °	145 °
Hip right	60 °	75 °	80 °	90 °	100 °	120 °	110 °
Hip left	75 °	85 °	95 °	100 °	110 °	115 °	120 °

To assess the patient's tissue oxygenation levels, we utilized the Train. Red FYER NIRS device, a continuous-wave NIRS sensor.[7] This device features a light-emitting diode that emits light at two wavelengths: 760 nm and 850 nm, and is equipped with a 64-pixel receiver positioned at a mid-range distance of 35 mm, known as the interpod distance. The sensor connects to a mobile app for data collection and transmission, and it is secured to the patient's quadriceps with a specially designed strap. The strap, made from flexible fabric, includes four non-slip silicone strips to ensure stability and prevent slipping down the leg (Table 3), (Figure2).

Figure 2. Quadriceps evaluation with NIRS technology

Intervention

Proposed plan of care

The patient received information regarding treatment methods for lymphedema management, as well as the necessary duration and precautions to minimize edema and lymphorrhea.[8] The objectives of the intervention were as follows: -to achieve a reduction in total limb circumference by 30 to 35 cm for the left leg and 20 cm for the right leg; -to improve skin texture, reduce fibrosis, and improve tissue health;-to reduce the risk of infection;-to facilitate independence in self-management

Table 3. Quadriceps evaluation with NIRS technology

		Week 0	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6
SmO_2 (%)	Right	63.25±11.83	65.43±11.83	65.28±11.82	66.51±11.83	66.51±11.83	67.73±11.55	68.53±11.09
	left	62.31±11.83	63.45±11.82	64.32±10.02	64.23±09.33	64.23±12.54	65.12±11.82	67.25±11.82
Hb Diff (µM)	Right	20.26±5.32	21.16±5.78	21.17±5.32	22.77±5.32	22.26±5.32	23.47±4.06	24.33±4.94
	left	21.02±4.94	22.17±4.06	23.47±4.06	23.01±4.32	23.34±4.06	24.02±4.94	24.65±4.94
O_2Hb (µM)	Right	62.33±11.21	63.33±12.65	63.33±12.24	64.22±11.76	63.33±12.66	64.33±12.24	65.33±12.89
	Left	63.87±13.18	6287±12.71	62.87±12.58	63.12±12.47	63.87±11.85	65.17±11.88	66.87±12.19

, including clothing use and care. The patient accepted the proposed care plan, and subsequent appointments were scheduled five times a week for 6 weeks.

Initiation of lymphedema management (complete decongestive therapy)

The patient underwent a comprehensive Complex Decongestive Physiotherapy (CDP) treatment protocol. The procedure involved 45 minutes of manual lymphatic drainage (MLD) following a proximal-to-distal and then proximal sequence. It also included lower-extremity skin care and hydration to preserve tissue integrity, as well as the application of multilayer short-stretch elastic bandages to both legs. This was achieved using cotton padding and foam layers to optimize pressure gradients. After bandaging, the patient performed a brief therapeutic exercise routine consisting of active ankle pumps, gentle lower-limb mobility movements, and diaphragmatic breathing to enhance the muscle-joint pump effect and support lymphatic flow.The CDT sessions were administered once daily, five days per week, over the course of the treatment period to ensure adequate decongestion and maintain the physiological effects of the therapy.

This treatment regimen was conducted five times a week over the course of six weeks. The therapy was provided by a physiotherapist certified in Manual Lymphatic Drainage Combined Decongestive Physiotherapy through the Turkish Lymphedema Association, who had more than seven years of experience treating lymphedema patients. The effectiveness of the treatment was assessed through quantitative measurements of leg edema, subjective reports of pain, range of motion, and quadriceps muscle oxygenation levels.

All measurements were quantitatively assessed by measuring the circumference of each leg before and after each treatment. After the first week, the patient experienced an improvement in the range of motion in her knees, particularly in her left knee, which had previously been more painful. Following the second week of treatment, as the swelling in her foot diminished, she was able to comfortably wear shoes. Normal joint movements continued in both an active and assistive manner. By week 3, the patient began walking 400 meters and gradually increased her distance to 1 kilometer. However, the 1-kilometer walk resulted in ankle pain, which subsided over the following week. The lymphorrhea episodes decreased substantially, and skin integrity improved. In week 4, her knee range of motion exceeded 120 degrees, leading to an increased walking pace. Her edema decreased, and muscle oxygenation increased as of this week In week 5, her hip range of motion also surpassed 120 degrees, enabling her to perform movements independently and with ease.

Outcome

By week 6, her range of motion had returned to normal limits in both the knee and hip, allowing her to walk 1 kilometer without any issues and at her desired pace. She also reported significant improvement regarding excessive sweating and groin pain, which had alleviated since her first treatment. Throughout the case study, the patient's outlook became increasingly optimistic, motivating her to engage in more physical activity. The patient underwent six weeks of decongestive lymphedema therapy and exceeded all therapeutic goals for limb reduction. Following treatment, she exhibited minimal fibrosis and reported a 12 kg weight loss, which allowed her to wear properly fitting clothes (Figure 3).

The lymphedema treatment proved to be effective, as the patient noted a significant reduction in the size of her lower limbs and expressed great satisfaction with the results. Ongoing care will be essential to maintain a continued reduction in limb size. Overall, decongestive lymphedema therapy was successful in addressing this patient’s primary lymphedema in the lower extremities.

Figure 3. The patient's legs after treatment

Discussion

This case presentation demonstrates the efficacy of rehabilitation methods such as manual lymphatic drainage and multi-layer compression bandaging in the treatment of advanced congenital lymphoedema complicated by lymphedema. This intervention resulted in a significant reduction in leg oedema volume, increased range of motion, improved muscle oxygenation as assessed by near-infrared spectroscopy (NIRS), and improvements in functional capacity over a six-week period. When compared with the current literature, these findings are consistent with studies reporting the effects of CDT on oedema control, muscle oxygen saturation, and functional gains in rare congenital lymphoedema.

In particular, the gradual alleviation of symptoms despite the patient's history of untreated lymphoedema demonstrates the potential for reversing morbidity even in chronic cases. The chronic accumulation of protein rich interstitial fluid, a prominent feature of primary lymphoedema, causes decreased muscle oxygenation and tissue remodelling, leading to reduced limb mobility and increased susceptibility to infection.[8] The patient's complaints of severe pain, knee and hip flexion impairment, and oedema are consistent with these known pathophysiological effects. The data in this case are similar to the case in the study by Greene et al.

Recent studies have shown that not only MLD but also holistic approaches are prominent, with conservative treatments accelerating lymphatic mobility and reducing limb volume and circumference. Furthermore, by activating the lymphatic and neural systems, they can facilitate tissue oxygenation by reducing sympathetic tone and increasing parasympathetic activity. Furthermore, the combined application of multi-layered, elastic bandages has demonstrated a significant ability to support volume reduction. This is achieved by mechanically supporting the natural flow of lymphatic fluid, facilitating optimal drainage, and preventing excessive fluid accumulation in tissues. The bandage's special design aids stabilisation and gentle compression, improving circulation and fluid elimination in the lymphatic system.[8-10]

A comprehensive review of the current literature reveals a substantial amount of research confirming the effectiveness of Manual Lymphatic Drainage (MLD) in various patient populations. Bilancini et al. found that patients with lymphoedema showed a marked decrease in lymphatic drainage compared to healthy individuals and therefore suggested that MLD could lead to a reduction in limb circumference and overall volume by facilitating lymphatic drainage.[7] Furthermore, findings reported by Marss emphasise the preventive role of MLD against haematoma formation and also demonstrate that it stimulates lymphangimotor activity in patients diagnosed with lipoedema.[11] Moreover, Schneider's research further confirms the multifaceted benefits that lymphoedema patients can derive from CDP interventions.[12] In our clinical observations following CDP application, we identified significant reductions in both limb volume and circumference, reinforcing the idea that CDP may play an important role in the therapeutic management of limb volume in affected patients. There are a limited number of studies investigating the short-term effects of various interventions on oxygenation of the quadriceps muscle. For example, one study found that 30 minutes of manual lymphatic drainage led to a significant increase in oxygen levels in the gastrocnemius muscle from 0.54±0.02 µmol/L to 0.69±0.02 µmol/L.[12] The results of our study are consistent with these findings.

When interpreting the results obtained in this case, it is important to consider the patient's 12 kg weight loss during the rehabilitation process. The reduction in body weight may have had positive effects on joint range of motion, pain perception, and functional capacity by reducing the mechanical load on the lower extremities.[13] However, it cannot be determined in this case presentation whether the weight loss was a direct result of the rehabilitation programme or accompanied lifestyle changes. Therefore, clinical improvements should be evaluated within the context of the combined effect of weight loss and the conservative rehabilitation approaches applied.

The rarity of isolated congenital lymphoedema and its association with lymphatic atresia increases the educational value of this case. The fact that positive clinical gains can be achieved even in a long-standing, advanced, and chronic case that has not received treatment for a long time provides an important clinical message highlighting the potential role of rehabilitation. However, due to the nature of case presentations, these findings cannot be directly generalised to general clinical practice. The results obtained are intended for clinical observation and hypothesis development rather than establishing causal relationships.

This case report demonstrates that a comprehensive rehabilitation program is effective in addressing the physiological, functional, and clinical needs of a patient with advanced congenital lymphedema requiring lymphatic drainage. The findings underscore the potential value of rehabilitation strategies in managing similar clinical conditions and provide a foundation for future larger, controlled studies.

Compliance with Ethical Standards

The study was conducted in accordance with ethical standards, ensuring that the patient’s privacy was fully protected. Written informed consent was obtained from the patient for the publication of this case report.

Conflict of Interest

The author declares that there is no potential conflict of interest.

Author contribition

EME; Performed the clinical evaluation of the case, conducted data collection, analysis, and interpretation,

carried out the literature review, designed the structure of the manuscript, and wrote the full.

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