OLGU SUNUMU CASE REPORT

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# Rehabilitation Approach in the Treatment of Advanced Lymphedema and Lymphorrhea: A Case Report and Review of the Literature

## İlerlemiş Lenfödem ve Lenforenin Tedavisinde Rehabilitasyon Yaklaşımı: Bir Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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**ABSTRACT** Lymphedema is a chronic condition that significantly reduces the quality of life and affects mobility and physical function. Although the complete decongestive therapy (CDT) includes manual lymph drainage, compression techniques, exercise, and skincare, additional approaches should be used when planning the treatment in case of lymphedema complications. In this report, a detailed evaluation of a patient with advanced lymphedema with lymphorrhea in the lower extremity and the methods applied in the treatment steps were explained. After the infection and lymphorrhea were treated with antibiotics and the four-layer bandage, the CDT was applied to the patient and leg diameters decreased distinctly.

**Keywords:** Complete decongestive therapy; compression therapy; four-layer bandaging; lymphoedema; lymphorrhea

**ÖZET** Lenfödem, yaşam kalitesini önemli ölçüde bozan, hareketliliği ve fiziksel işlevi etkileyen kronik bir durumdur. Komplet dekonjestif tedavi (KDT) manuel lenf drenajı, kompresyon teknikleri, egzersiz ve cilt bakımını içermektedir fakat komplike lenfödemde tedavi planlanırken ek yaklaşımlardan da yararlanılmalıdır. Bu olgu sunumunda, alt ekstremitesinde lenfore mevcut ileri evre lenfödemli bir hastanın detaylı değerlendirmesi ve tedavi basamaklarında uygulanan yöntemler anlatılmıştır. Enfeksiyon ve lenfore antibiyotik ve 4 katmanlı bandaj ile tedavi edildikten sonra hastaya KDT uygulanmış ve tedavi sonucunda bacak çaplarında belirgin azalma gözlenmiştir.

**Anahtar Kelimeler:** Komplet dekonjestif tedavi; kompresyon tedavisi; dört katmanlı bandaj; lenfödem; lenfore

Lymphoedema is a progressive, often-debilitating disease, caused by intrinsic or acquired defects related to localized or general protein-rich fluid accumulation, resulting from congenital malformation or impairment.[1,2] It impairs the patient physically and psychologically, reducing the quality of life. There are two types of lymphedema: Primary lymphedema and secondary lymphedema. Primary lymphedema occurs because of congenital anomalies in the lymphatic system. Secondary lymphedema however develops due to several reasons including trauma, surgical intervention, radiotherapy, infection, or obesity. Noncancerous lymphedema usually cannot be noticed until it has progressed, therefore making it significantly harder to manage the disease.[2] The conservative approach to the treatment of lymphedema includes some complex physical therapy methods such as lymphatic drainage and compression

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treatments by using elastic bandages or elastic stockings.[3] Among the bandaging methods, multi-layer bandaging is often used in the treatment of edema and is applied to the entire lower extremity while four-layer bandaging is applied in venous leg ulcers and remains limited to the cruris.[4]

In this case report, we aimed to employ a multidisciplinary approach and present the steps of the complex physical therapy program applied in the treatment of a patient with advanced lymphoedema accompanied by lymphorrhea.

## CASE REPORT

A morbid obese female patient referred to our outpatient clinic due to swollen leg and increasing complaints over about 10 days where she was diagnosed of Stage 3 lymphedema (according to International Lymphology Society classification) in her both lower extremities. The patient's lymphedema which covered an area from the distal part to the thigh spread to the abdominal area. There was discoloration on the skin of the patient, pink-brown bullous lesions, and it was accompanied by pronounced lymphorrhea. The range of the lower extremity motion was limited, and therefore the patient has to be mobilized in a wheelchair (Figure 1). Additionally, the patient was diagnosed with Type 2 diabetes mellitus, hypothyroidism, and chronic obstructive pulmonary disease. The patient complained about difficulty in breathing due to her hypervolemic condition. She had a decubitus ulcer on her left heel. The arterial and venous flows were reported as normal in the doppler ultrasonography. In medical laboratory tests, there was a worsening in kidney function of the patient (creatinine level 3 mg/dL). Before planning the rehabilitation, clinical consultations of relevant clinics were gathered. Clinical opinion was obtained from the nephrology clinic due to the worsening of renal function. Also, consultations were made with the infectious disease clinic for cellulitis and lymphorrhea, the chest disease clinic due to dispnea, and the cardiology clinic for possible heart problems. In the thorax computed tomography scanning, significant effusion was detected in the lung. There was no cardiological pathology. Also, there was an increase in erythrocyte sedimentation rate and C-reactive protein levels. Consequently, during the first week, the patient started dialysis after nephrology consultation in addition to antibiotic treatment for cellulitis and lymphorrhea. Empirical teicoplanin and piperacillin treatment were started for ulcerous lesions and lymphorrhea in both legs. After the first 7 days of the regular antibiotherapy and dialysis, there was a significant decrease in the amount of lymphorrhea and the patient's general condition improved. Also, dyspneic breathing lessened. A rehabilitation program including bandaging treatment was started for the patient in the 2nd week. A four-layer bandage set, which was changed every 3-day for a period of 6-week, was applied to the patient by a physiotherapist who had training and experience in lymphedema (Figure 2).

Consent for Publication Written informed consent was obtained from the patient for publication of this case report.

**FIGURE 1:** Lymphedema, cellulitis, and lymphorrhea in bilateral legs and edema in abdominal region at baseline.

**FİGURE 2:** Application of the four-layer bandage.

Before bandaging, the skin was wiped with isotonic water and urea and Cutimed® PROTECT (BSN Medical, Essity, Sweden) protect were applied. Piperacillin treatment was stopped after the detection of pseudomonas in the biopsy sample taken from the left heel, and meropenem was started. Dialysis frequency decreased due to improvement in kidney function tests and an appropriate dose of furosemide was started to be given to prevent hypervolemia. Between the 7th and 11th weeks of compression therapy, a four-layer bandage treatment was maintained as the heel wound in the left lower extremity did not improve, and a compression bandage was applied to the right lower extremity (Figure 3 a,b). While the patient was bedridden in the first weeks, ten weeks later she started therapeutic exercise in the physical therapy unit where she was accompanied by a supervisor. Between the 11th and 12th weeks, compression bandage, manual lymph drainage, and exercise therapy were implemented for both lower extremities. Complete decongestive therapy (CDT) was applied 5 days per week. Improvement in kidney function tests was observed in the following days and the dialysis treatment was stopped. The antibiotic treatment was also discontinued after a decrease in acute phase reactants. She was able to start walking independently to the physical therapy unit. At the end of the 13th week, the patient who received a total of 54 sessions of physical therapy was able to mobilize independently (Figure 4 a,b). The patient was discharged with a suitable compression garment and hygienic self-care was recommended. Throughout 13-week lymphedema rehabilitation, an average of 15 cm reduction in circumferential measurements of both lower extremities as well as an improvement in leg volumes were achieved, and a weight loss of 27 kg was observed (Table 1). Abdominal edema and lymphorrhea also disappeared.


**FIGURE 3:** a) After 7-week of compression therapy, b) Short tension bandage applied to the right leg, the four-layer bandage continued on the left leg.


**FIGURE 4:** a) After 13-week compression therapy, b) Short tension bandage applied to both legs.

## DISCUSSION

In this case report, the rehabilitation process of advanced-stage lymphedema patient with lymphorrhea, renal failure, and secondary bacterial infection was described in detail. For patients with a diagnosis of advanced lymphedema and lymphorrhea, we suggest application of four-layer bandage until ulcer or skin condition improves, which should also be followed by the lymphatic drainage and multi-layer bandaging treatment, known as CDT. Also, the systemic evaluation of the patients should not be skipped, and consultations should be gathered from different disciplines when necessary.

The conservative treatment for lymphedema consists of physical therapy, medication, and psychosocial rehabilitation. Physical therapy includes CDT, intermittent pneumatic compression, laser, pressure garment, and specific exercises. Commonly anti-microbials (in case of acute inflammation such as cellulitis, lymphangitis or erysipelas), calcium dobesilate, and diosmin-hesperidin can be used as drug therapy in patients with lower extremity edema.[5-7] The main treatment for lymphedema is based on CDT (multi-layer short stretch bandage, manual lymph drainage, skincare, and exercises). While it causes a reduction in the increasing volume due to lymphedema in the first phase of the treatment, preservation of volume reduction in the second phase is also important. Multi-layer short stretch bandage and elastic compression garments are the basic elements of complete decongestive physiotherapy.[8-10] Different treatments have also been tried for patients with complicated and advanced lymphedema. There is a case report showing that the use of a different adaptive velcro compression system (Juxta-fit, Medi UK) proved effective in the treatment of advanced lymphoedema and this method was suggested to be used as an alternative to compression bandage.[2] Also, there are some case reports and case series in the literature examining the implementation of multi-layer short stretch compression and diuretic treatments (mostly furosemide).[10-12] In a case series, compression therapy combined with a diuretic drug (furosemide) has been found to be beneficial in the patients with refractory advanced lymphedema.[11]

According to the consensus report of the International Journal of Lymphology, although diuretic therapy can be used as an adjunct to treatment in the initial phase of CDT, its long-term use is not recommended as it may cause fluid/electrolyte imbalance and may induce chronicity of edema due to disruption of the

**TABLE 1:** Comparison of the lower extremity dimensions before and after the treatment.

| | Before the treatment | | After the treatment | | The difference | |
|---|---|---|---|---|---|---|
| | Left | Right | Left | Right | Left | Right |
| 1st MTP (cm) | 30.5 | NE | 24.2 | 24.3 | 6.3 | NE |
| Ankle (4 cm) | 40.7 | 40.1 | 24.0 | 23.2 | 16.7 | 16.9 |
| 8 cm | 46.5 | 46.0 | 27.4 | 26.2 | 19.1 | 19.8 |
| 12 cm | 52.5 | 54.0 | 31.5 | 31.0 | 21.0 | 23.0 |
| 16 cm | 56.5 | 59.4 | 36.9 | 36.2 | 19.6 | 23.2 |
| 20 cm | 62.0 | 61.5 | 40.8 | 40.1 | 21.2 | 21.4 |
| 24 cm | 63.9 | 61.4 | 43.5 | 42.7 | 20.4 | 18.7 |
| 28 cm | 63.4 | 59.4 | 44.6 | 43.4 | 18.8 | 16.0 |
| 32 cm | 56.1 | 54.6 | 44.4 | 44.0 | 11.7 | 10.2 |
| 36 cm | 57.8 | 56.6 | 44.5 | 44.1 | 13.3 | 12.5 |
| 40 cm | 59.9 | 59.1 | 46.5 | 45.8 | 13.4 | 13.3 |
| 44 cm | 65.0 | 63.2 | 52.8 | 52.2 | 12.2 | 11.0 |
| 48 cm | 68.4 | 67.8 | 56.0 | 54.8 | 12.4 | 13.0 |
| 52 cm | 69.8 | 69.3 | 58.9 | 57.8 | 10.9 | 11.5 |
| 56 cm | 71.3 | 70.9 | 60.5 | 60.2 | 10.8 | 10.7 |
| Limb volume (mls) | 15.038 | 14.634 | 8.356 | 8.078 | 6.682 | 6.562 |

MTP: Metatarsophalangeal joint; NE: Not evaluated.

renin-angiotensin system.[10,13] In some selected cases where conservative treatment was not effective, surgical treatment methods such as debulking, liposuction, and microsurgical procedures were applied.[5]

Obstruction or malformation of lymphatic vessels leads to the accumulation of fluid, waste products, and immune mediators. The accumulation of these mediators triggers an irregular lymphatic flow and abnormal immune cell activation cycle, thereby creating a difficulty in wound healing.[14] Venous leg ulcer is known to be the most common comorbidity in the lymphedema patients (non-cancerous). And 18% of patients with venous leg ulcers do not receive any treatment for lymphedema.[15] In a Cochrane review, it was reported that the most effective compression bandage technique in the treatment of ulcer is four-layer bandaging.[16] Also, continuous negative pressure wound therapy and Unna boot which are applied in addition to compression bandage for lymphedematous leg with infected decubitus ulcers have been found to be effective.[14,17] In our case, before the CDT, four-layer bandaging technique (1 orthopedic wool bandage, 1 light support bandage, 1 light compression bandage, 1 cohesive extensible bandage) was employed to cure the accompanying cellulitis and infection. Multilayer bandaging is one of the steps of CDT and cellulite constitutes a contraindication for this treatment.[4] Therefore, the skin wounds of the patient healed in the 11th week thanks to antibiotherapy, hygienic skincare, and four-layer bandaging method, and then CDT was started for both legs.

Consequently, complications due to lymphedema may put the patients in serious life-threatening conditions. Education and training of the physicians about the complications and management of lymphedema to plan the most appropriate treatment program with a multidisciplinary approach when needed is essential. Examination of the patients and prompt implementation of the suitable treatment methods are of utmost importance in therapeutic response.

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### *Conflict of Interest*

*No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.*

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