

# Lipedema in Male Progressing to Subclinical and Clinical Systemic Lymphedema

Livia Maria Pereira de Godoy<sup>a, b</sup>, Maria de Fatima Guerreiro Godoy<sup>b</sup>, Jose Maria Pereira de Godoy<sup>b, c, d, e</sup>

## Abstract

The aim of the present case study was to report the 3-year follow-up of a male patient with lipedema and subclinical systemic lymphedema evaluated using multi-segment bioimpedance. The report describes the case of a 53-year-old male with a history of oncological surgery involving lymph node clearance in the right inguinal region followed by radiotherapy and chemotherapy. The physical examination revealed lipedema and lymphedema in the right lower limb. The patient was submitted to multi-segment bioimpedance, circumference measurements and volumetry, with the detection of clinical lymphedema of the limb. The patient underwent intensive treatment for lymphedema using the Godoy Method®, which resulted in a substantial reduction in the edema. However, at the 1-year follow-up, the patient had progressed to subclinical systemic lymphedema, followed a year later by clinical systemic lymphedema evaluated using multi-segment bioimpedance. Lipedema is less frequent in men compared to women, but the increase in weight is an aggravating factor in both sexes. This condition initially affects the lower limbs, progressing to subclinical systemic lymphedema, followed by clinical systemic lymphedema determined using multi-segment bioimpedance, demonstrating that edema in patients with lipedema may be systemic.

**Keywords:** Lipedema syndrome; Male sex; Lymphedema; Subclinical; Clinical systemic lymphedema

#### Introduction

Lipedema is a clinical condition that affects approximately 10% of women, not described in literature in male, and is characterized by a greater distribution of fat in the extremities [1-

Manuscript submitted February 7, 2022, accepted May 16, 2022 Published online June 2, 2022

<sup>d</sup>Research CNPq (National Council for Research and Development), Brazil <sup>e</sup>Corresponding Author: Jose Maria Pereira de Godoy, Floriano Peixoto 2950, Sao Jose do Rio Preto, SP, Brazil. Email: godoyjmp@gmail.com

doi: https://doi.org/10.14740/jmc3806

3]. The redistribution of fat tissue is the main clinical aspect, but some physiopathological processes may be detected over time. Immunohistochemistry shows degenerative and regenerative changes of the lipedema tissue, characterized by crownlike structures and proliferation of adipose tissue, suggesting increased adipogenesis tissue [3-6]. Changes in microcirculation, specifically in the lymphatic system, are the most prominent anatomical and functional findings [7, 8].

A study characterized in detail morphologic and molecular alterations in the adipose tissue composition of lipedema patients compared with healthy controls in histological analysis of the infiltrate of immune cells confirmed the increased presence of macrophages, with no changes in the T-cell compartment, increase in intracellular fibrosis or adipocyte hypertrophy [9]. Obesity is an associated comorbidity in 80% of patients with lipolymphedema [10].

A study involving magnetic resonance lymphography (without contrast) in patients with lipedema that progressed to lipolymphedema of the lower limbs detected dilated peripheral lymphatic vessels, indicating a subclinical state of lymphedema [11]. Lymphatic vessels begin to dilate and elongate with the progression of lipedema, leading to the development of microaneurysms [7, 12].

A recent study involving multi-segment bioelectrical impedance analysis found that morbidly obese patients developed subclinical systemic lymphedema that can progress to clinical systemic lymphedema, which is aggravated by an increase in the body mass index (BMI) [13-19]. Individuals with lipedema develop these conditions in earlier stages and lymphedema is detected by multi-segment bioimpedance in 50% of those with a BMI higher than 30 kg/m<sup>2</sup>.

The aim of the present study was to report the 3-year follow-up of a male patient with lipedema and subclinical systemic lymphedema evaluated using multi-segment bioimpedance.

## **Case Report**

#### Investigations

A 53-year-old male patient with a history of oncological surgery, prostate, involving lymph node clearance in the right inguinal region followed by radiotherapy and chemotherapy subsequently developed edema of the right lower limb and

Articles © The authors | Journal compilation © J Med Cases and Elmer Press Inc™ | www.journalmc.org This article is distributed under the terms of the Creative Commons Attribution Non-Commercial 4.0 International License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited

<sup>&</sup>lt;sup>a</sup>Dermatology in Instituto Lauro de Souza Lima, Bauru, Brazil <sup>b</sup>Researcher Group of the Clinica Godoy, Sao Jose do Rio Preto, SP, Brazil <sup>c</sup>Cardiology and Cardiovascular Surgery Department of the Medicine School in Sao Jose do Rio Preto, FAMERP, Brazil

	Total (mL)	Normal water values (mL)	Total extracellular water/to- tal body water ratio (mL)
Total intracellular water	36.4	26.5 - 32.5	
Total extracellular water	23.7	16.3 - 19.9	
Total extracellular water/total body water ratio	0.395		0.390
Right arm	3.251	2.56 - 3.12	0.382 limit (0.36 - 0.39)
Left arm	3.10	2.56 - 3.12	0.385 limit (0.36 - 0.39)
Trunk	24.2	20.4 - 24.9	0.389 limit (0.36 - 0.39)
Right leg	14.81	7.10 - 8.68	0.421 limit (0.36 - 0.39)
Left leg	9.86	7.10 - 8.68	0.380 limit (0.36 - 0.39)

Table 1. Body Liquids at First Evaluation Determined by Multi-Segment Bioimpedance

sought a specialized service for the treatment of lymphedema. The physical examination revealed edema from the root of the thigh involving the entire limb. The limb had the physical appearance of lipedema, for which the patient had a family history. At the first evaluation by bioimpedance, the patient's BMI was 29 kg/m<sup>2</sup>.

## Diagnosis

Circumference measurements, volumetry and multi-segment bioimpedance were performed for diagnosis and evaluation treatment. The diagnosis of lipedema is primarily clinical based on history and physical examination. Multi-segment bioimpedance is essential to diagnose systemic edema that suggests changes in the lymphatic system. It is fundamental in the evolution of these patients and was performed during his follow-up period.

## Treatment

The treatment for lymphedema first time in the Clinica Godoy, Sao Jose do Rio Preto, Brazil was during 5 days (8 h per day), involving associated therapy, Godoy method<sup>®</sup>: cervical lymphatic therapy, mechanical lymphatic therapy (RAGodoy<sup>®</sup>) and grosgrain stocking. Maintenance with grosgrain stocking and mechanical lymphatic therapy returns one time per year. Lipedema has pathophysiological changes related to the lymphatic system and the same treatment of lymphedema is performed in lipedema [18].

## Follow-up and outcomes

Below the knee, the right limb had a volume of 4,720 g and the left limb had a volume of 3,419 g (difference between limbs: 1,224 g). During the 3-year follow-up of the case, only a reduction to 4,100 g in the right leg was maintained. Multisegment bioimpedance showed the progression from subclinical systemic lymphedema to clinical systemic lymphedema throughout the 3-year follow-up, as displayed in Tables 1, 2 and 3. At the first evaluation, abnormally high intracellular and extracellular water, total body water as well as water in both lower limbs and one upper limb were found. Lymphedema detected by bioimpedance was found in the right leg. At the second evaluation, the patient had progressed to subclinical systemic lymphedema, with more water in all extremities and the trunk and bilateral lower limb lymphedema detected by bioimpedance. Three years after the original evaluation, multi-segment bioimpedance revealed clinical systemic lymphedema, with the increase in water in all extremities. The patient was instructed to lose weight and maintain treatment for lymphedema.

Table 2. Body Liquids at Second Evaluation (1 Year After First Evaluation) Determined by Multi-Segment Bioimpedance

	Total (mL)	Normal water values (mL)	Total extracellular water/to- tal body water ratio (mL)
Total intracellular water	39.6	26.5 - 32.5	
Total extracellular water	25.6	16.3 - 19.9	
Total extracellular water/total body water ratio	0.394		0.390
Right arm	3.94	2.56 - 3.12	0.372 limit (0.36 - 0.39)
Left arm	3.84	2.56 - 3.12	0.382 limit (0.36 - 0.39)
Trunk	28.1	20.4 - 24.9	0.389 limit (0.36 - 0.39)
Right leg	13.58	7.10 - 8.68	0.413 limit (0.36 - 0.39)
Left leg	10.37	7.10 - 8.68	0.393 limit (0.36 - 0.39)

	Total (mL)	Normal water values (mL)	Total extracellular water/to- tal body water ratio (mL)
Total intracellular water	35.5	26.5 - 32.5	
Total extracellular water	23.6	16.3 - 19.9	
Total extracellular water/total body water ratio	0.400		0.390
Right arm	3.41	2.56 - 3.12	0.390 limit (0.36 - 0.39)
Left arm	3.36	2.56 - 3.12	0.391 limit (0.36 - 0.39)
Trunk	25.4	20.4 - 24.9	0.395 limit (0.36 - 0.39)
Right leg	13.54	7.10 - 8.68	0.417 limit (0.36 - 0.39)
Left leg	9.64	7.10 - 8.68	0.398 limit (0.36 - 0.39)

Table 3. Body Liquids at Third Evaluation (3 Years After First Evaluation) Determined by Multi-Segment Bioimpedance

## Discussion

This paper reports the first case of the follow-up of lymphedema/lipedema evaluated by multi-segment bioimpedance in a male patient. The increase in intracellular and extracellular water as well as water in the extremities initially called attention to this condition, but there was no knowledge regarding the evolution of such cases. At the second evaluation 1 year later, the patient presented subclinical systemic lymphedema, which progressed to clinical systemic lymphedema at the 3-year evaluation, as determined by multi-segment bioimpedance.

We have been following patients with lipedema and lymphedema for approximately 8 years, after having included multi-segment bioimpedance in the evaluation of patients, but this is the first case of lipedema progressing to systemic lymphedema in a male patient. Obesity is the main aggravating factor of edema in these patients and the prevalence of subclinical systemic lymphedema increases with the increase in BMI.

With regard to the physiopathology of lipedema, the main changes in which we can interfere involve the lymphatic system [18]. Using multi-segment bioimpedance, we have found that obesity is a major aggravating factor, as a higher BMI translates to greater lymphatic harm. Therefore, the main form of treatment for such patients is to address obesity through weight reduction, which is the only way we currently know to reverse this lymphedema and achieve standards of normality or near normality. Patients with lymphedema and obesity who lose weight experience an important reduction in the edema and, without an additional harm to the lymphatic system, can achieve normality [19, 20].

Lipedema is less frequent in men compared to women, but the increase in weight is an aggravating factor in both sexes. This condition initially affects the lower limbs, progressing to subclinical systemic lymphedema, followed by clinical systemic lymphedema determined using multi-segment bioimpedance, demonstrating that edema in patients with lipedema may be systemic.

Lipedema in women may have other aggravating factors such as idiopathic cyclic edema and esthetic cellulitis [21, 22]. In men, these alterations were not identified. However, it is essential to identify in each patient with lipedema all aggravating factors of the edema and treat each of the pathophysiological aspects involved. However, weight gain is the most serious of all and these are factors that can interfere. Other alterations are described in lipedema of genetic causes, but we cannot interfere. At the moment, weight loss and lymphatic therapy to reduce edema are the main therapeutic bases.

In around 10% of women, the physical aspect of lipedema is detected, but rare in literature in men; as long as they remain thin, the appearance of edema and symptoms is less frequent, a fact that characterizes the lipedema syndrome. Therefore, the patient in the present study has lymphedema and lipedema syndrome [1].

#### Learning points

Lipedema is less frequent in men compared to women, affects the lower limbs, and can progress to subclinical systemic lymphedema.

Weight gain is one of the most aggravating factors of lipedema.

Lipedema in women may have other aggravating factors such as idiopathic cyclic edema and esthetic cellulitis, but in men, these alterations were not identified.

## Acknowledgments

None to declare.

## **Financial Disclosure**

The authors declare no financial support or funding for study.

## **Conflict Interest**

The authors declare no conflict of interest.

## **Informed Consent**

Patient's informed consent for publication of this report was obtained.

# **Author Contributions**

JMP Godoy: design, analysis and interpretation of data, data collection, article writing, critical review of the text, statistical analysis, and overall responsibility for the study. MFG Godoy and LMP Godoy: analysis and interpretation of data, data collection, article writing, critical review of the text, statistical analysis, and overall responsibility for the study. All authors read and approved the final manuscript.

# **Data Availability**

The authors declare that data supporting the findings of this study are available within the article.

# References

- 1. Wold LE, Hines EA, Jr., Allen EV. Lipedema of the legs; a syndrome characterized by fat legs and edema. Ann Intern Med. 1951;34(5):1243-1250.
- Redondo Galan C, Garcia Bascones M, Marquina Valero MA. [Lipoedema: Symptoms, diagnosis and treatment. A literature review]. Rehabilitacion (Madr). 2019;53(2):104-110.
- 3. Herbst KL, Mirkovskaya L, Bharhagava A, Chava Y, CHT Te. Lipedema fat and signs and symptoms of illness, increase with advancing stage. Arch Med. 2015;7(4):10.
- 4. Suga H, Araki J, Aoi N, Kato H, Higashino T, Yoshimura K. Adipose tissue remodeling in lipedema: adipocyte death and concurrent regeneration. J Cutan Pathol. 2009;36(12):1293-1298.
- Buso G, Depairon M, Tomson D, Raffoul W, Vettor R, Mazzolai L. Lipedema: a call to action! Obesity (Silver Spring). 2019;27(10):1567-1576.
- 6. Pereira de Godoy LM, Pereira de Godoy HJ, Pereira de Godoy Capeletto P, Guerreiro Godoy MF, Pereira de Godoy JM. Lipedema and the evolution to lymphedema with the progression of obesity. Cureus. 2020;12(12):e11854.
- Amann-Vesti BR, Franzeck UK, Bollinger A. Microlymphatic aneurysms in patients with lipedema. Lymphology. 2001;34(4):170-175.
- Al-Ghadban S, Cromer W, Allen M, Ussery C, Badowski M, Harris D, Herbst KL. Dilated blood and lymphatic Microvessels, angiogenesis, increased macrophages, and adipocyte hypertrophy in lipedema thigh skin and fat tissue. J Obes. 2019;2019:8747461.

- 9. Felmerer G, Stylianaki A, Hagerling R, Wang A, Strobel P, Hollmen M, Lindenblatt N, et al. Adipose tissue hypertrophy, an aberrant biochemical profile and distinct gene expression in lipedema. J Surg Res. 2020;253:294-303.
- 10. Herpertz U. Der Mibrauch des Lipodems. Lymphologie in Forschung und Praxis. 2003;7:90-93.
- 11. Cellina M, Gibelli D, Soresina M, Menozzi A, Martinenghi C, Panzeri M, Oliva G. Non-contrast MR Lymphography of lipedema of the lower extremities. Magn Reson Imaging. 2020;71:115-124.
- Bilancini S, Lucchi M, Tucci S, Eleuteri P. Functional lymphatic alterations in patients suffering from lipedema. Angiology. 1995;46(4):333-339.
- 13. de Godoy JMP. Systemic subclinical lymphedema due to obesity as the cause of clinical lymphedema: A new concept. Med Hypotheses. 2019;131:109312.
- Godoy MFG, de Godoy JMP, de Godoy LMP, de Godoy HJP, Ramos RR. Intensive treatment of lymphedema and mobilization of liquids in body segments. Medical Science. 2021; 25(113):1578-1582.
- 15. de Godoy JMP, de Godoy HJP, de Godoy ACP, Godoy MFG. Subclinical systemic lymphedema in patients with obesity and lipedema Clin Pract. 2019;16(SI):1289-1291.
- 16. de Godoy JMP. Diagnostic criteria and clinical evolution of systemic lymphedema caused by obesity: bioimpedance analysis. Ann Med Health Sci Res. 2019;9:420-421.
- 17. de Fatima Guerreiro Godoy M, Silva EB, de Godoy JM. Bioimpedance to screen for abdominal fat in patients with breast cancer treatment-related lymphedema. Breast Dis. 2016;36(2-3):73-76.
- 18. Pereira de Godoy JM, Guerreiro Godoy MF. Hypotheses and evolution in the current treatment of lipedema syndrome. J Clin Med Res. 2022;14(2):106-107.
- 19. Pereira de Godoy JM, Pereira de Godoy LM, Pereira de Godoy AC, Guerreiro Godoy MF. Bariatric surgery and the evaluation of subclinical systemic lymphedema. J Surg Case Rep. 2019;2019(2):rjz028.
- 20. de-Godoy JMP, de-Godoy HJP, de-Godoy ACP, Godoy MFG. Obesity and systemic lymphedema: new concept evaluated by multisegmental bioimpedance analysis. Medical Science. 2021;25(112):1269-1271.
- 21. Pereira de Godoy JM, Pereira de Godoy HJ, Pereira de Godoy LM, Guerreiro Godoy MF. Prevalence of idiopathic cyclic edema in women with lower limb lymphedema. J Clin Med. 2017;7(1):2.
- 22. de Godoy JM, Barufi S, Godoy Mde F. Lipedema: is aesthetic cellulite an aggravating factor for limb perimeter? J Cutan Aesthet Surg. 2013;6(3):167-168.