ELSEVIER

Surgery for Obesity and Related Diseases ■ (2022) 1–6

SURGERY FOR OBESITY AND RELATED DISEASES

Original article

Persistent lipedema pain in patients after bariatric surgery: a case series of 13 patients

Manuel E. Cornely, M.D.[a,*], Till Hasenberg, M.D.[b], Oliver A. Cornely, M.D.[c,d,e], Christian Ure, M.D.[f], Christian Hettenhausen, Ph.D.[a], Jeremias Schmidt, M.D.[g]

[a]*Ly.Search GmbH, Koln, Germany*

[b]*Helios Obesity Center West, Helios St. Elisabeth Hospital Oberhausen, Witten/Herdecke University, Helios University Hospital Wuppertal, Wuppertal, Germany*

[c]*Department of Internal Medicine, University Hospital Cologne, Cologne, Germany*

[d]*Faculty of Medicine, University of Cologne, Cologne, Germany*

[e]*Translational Research, Cologne Excellence Cluster on Cellular Stress Responses in Aging-Associated Diseases, Cologne, Germany*

[f]*Department of Lymphology, Hospital Wolfsberg, Wolfsberg, Austria*

[g]*Department of Plastic, Aesthetic and Reconstructive Microsurgery/Hand Surgery, Hospital Ernst von Bergmann, Potsdam, Germany*

Received 7 July 2021; accepted 30 December 2021

Abstract

Background: Lipedema often remains undiagnosed in patients with obesity, leading to mismanagement of treatment. Because of this, despite remarkable weight loss after bariatric surgery and decreases in hip and abdomen circumference, some patients show only small decreases in circumference of the extremities and report persistent limb pain.

Objective: The goal of this work is to raise awareness of lipedema coincident with obesity, mistakenly diagnosed as obesity alone, in order to ensure the correct diagnosis of the condition and to achieve better treatment outcomes for people with lipedema and coincident obesity.

Setting: CG Lympha Clinic, Cologne, and Ernst von Bergmann Clinic, Potsdam.

Methods: From clinical records, we identified 13 patients who were diagnosed with lipedema only after undergoing bariatric surgery. We describe the course of their pain before and after bariatric surgery, focusing on the long-term progression of symptoms accompanying the disease.

Results: Lipedema cannot be cured by bariatric surgery, and although the patients in this study lost an average of more than 50 kg of weight, they displayed no improvement in the pain symptoms typical of lipedema.

Conclusions: Because of the different etiologies of lipedema and obesity, lipedema requires its own specific treatment. Patients suffering from obesity should always be assessed for pain and lipedema. If coincident lipedema is diagnosed, we suggest that bariatric surgery only be performed first if diet and exercise have failed, the patient's body mass index is >40 kg/m^2, and the patient has been informed of the possible persistence of pain. Lipedema, like a coincident disease, must be additionally treated conservatively or preferably surgically. This optimized treatment may help to better manage patient expectations after weight loss. (Surg Obes Relat Dis 2022;■:1–6.)

Keywords: Lipedema; Liposuction; Lipohyperplasia dolorosa; Obesity; Bariatric surgery

*Correspondence: Manuel E. Cornely, M.D., LY.SEARCH, Biberstrasse 7, 50678 Cologne, Germany.

E-mail address: post@lysearch.de (M.E. Cornely).

https://doi.org/10.1016/j.soard.2021.12.027

Lipedema was first described by Allen and Hines in 1940. It is a disproportionate, bilateral, and symmetrical accumulation of subcutaneous fat of the lower and occasionally upper extremities [1–3]. In lipedema, fat pads extend to the ankles and wrists, with the feet and hands spared, resulting in the characteristic cuff sign [4] (Fig. 1). Lipedema is a chronic and often progressive disease and mainly affects women, with an onset during puberty or at other times of hormonal change such as pregnancy or menopause [5]. The pathogenesis remains unclear, but it appears to be a genetic disorder with familial clustering and either X-linked dominant inheritance or, more likely, autosomal dominant inheritance with sex restriction [5]. Because of the lack of comprehensive studies on the prevalence of lipedema, there are no valid figures for affected women, but an incidence of 7%–9.7% is estimated in Germany [6,7].

Lipedema is classified into 3 stages based on its morphology (Fig. 2). Painfulness of the tissue does not correlate with the stage or extent of the disease affecting the limb [8]. In stage 1, the legs are slim but painful. In stage 2, the legs appear thicker or obese and are painful with an uneven, wavelike skin surface, and in stage 3, the legs are painful and often obese with bulky, drooping fatty tissue deformations [3,9]. The same applies to the arms in more than 90% of patients, although the disease often develops with a time delay of about 5 years after the legs. Because a reliable marker for lipedema is still missing [10,11], diagnosis is based on clinical examination, pain, and medical history only (Table 1).

The disease often coincides with elevated body mass index (BMI) and obesity [5,12,13]. Differentiation from other adipose tissue disorders can be challenging, and lipedema is frequently misdiagnosed as lifestyle-induced obesity or lymphedema [14]. In contrast to obesity, patients suffer from pain in the extremities, tenderness on palpation, and a tendency to bruise easily [2].

The fat in the arms and legs of affected people consists of hypertrophic and hyperplastic adipocytes [15–17]. While hypertrophic adipocytes decrease in size under diet and exercise or after bariatric surgery and reduction of obesity, physical reduction in the number of hyperplastic cells via liposuction has been shown to be of benefit in the treatment of lipedema [15,16,18]. However, because of a lack of awareness surrounding lipedema, the diagnosis and treatment of this disease are often delayed by years.

In our lipedema consultation, we have repeatedly encountered cases in which patients with obesity have undergone bariatric surgery without a prior lipedema diagnosis (Fig. 3). By summarizing these cases, we would like to draw attention to this problem and emphasize that lipedema pain in the legs cannot be reduced by bariatric surgery. Therefore, under certain circumstances, bariatric surgery should not be the first treatment following a diagnosis of coincident lipedema and obesity.

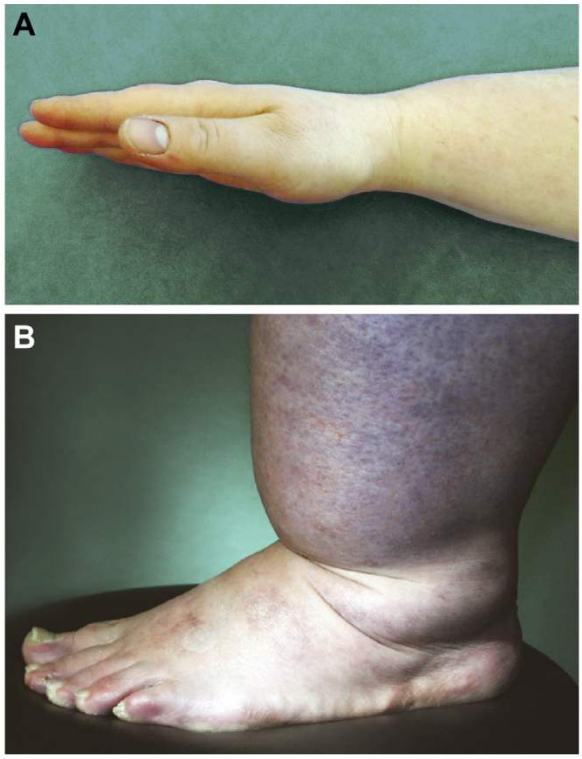

Fig. 1. Clinical picture of the characteristic cuff signs on arms (A) and legs (B).

Methods

In a retrospective analysis, we reviewed the medical records of all patients seen between January and October 2020 in 2 clinics in Germany specializing in the treatment of lipedema: the CG Lympha Clinic in Cologne and the Ernst von Bergmann Clinic in Potsdam. We focused on patients who had bariatric surgery prior to their diagnosis of lipedema. We documented the patients' current lipedema stage, height, weight, and leg pain and asked them about the severity of lipedema pain before bariatric surgery. For quantification of pain, we used the visual analog scale (VAS), whose scale ranges from 0 (= painless) to 10 (= very painful). We also asked the treating bariatric surgeons to report patients' height and weight from before the procedure. We documented and compiled all cases with complete data sets. Percentage excess weight loss was calculated using the following formula: percent excess weight loss = (weight before bariatric surgery − weight after bariatric surgery)/(weight before bariatric surgery − 25 × height^2) × 100. Percentage total weight loss was calculated according to the following formula: percent total weight loss = (weight before bariatric surgery − weight after bariatric surgery)/(weight before bariatric surgery/100). Data are reported as average ± standard deviation (SD). Pre- and postoperative differences within subjects were evaluated by a paired t test. Significance was set at $P < .05$.

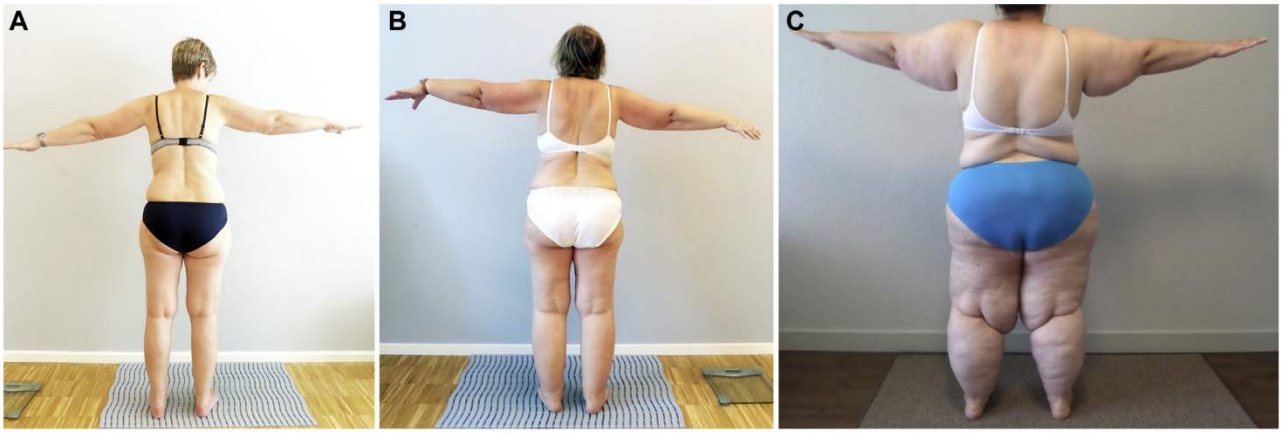

Fig. 2. Lipedema stage 1 (A), stage 2 (B), and stage 3 (C).

According to the data protection laws of North Rhine–Westphalia (Cologne) and Brandenburg (Potsdam), retrospective analysis and anonymized reporting of patient data without informed consent is appropriate, and ethics approval was waived. The data sets used and/or analyzed during this study are available from the corresponding author upon reasonable request.

Results

We identified 13 patients who suffered from lipedema-associated symptoms after bariatric surgery (Table 2). All patients reported a tendency to bruise; 9 of the 13 patients (69%) had a family history of lipedema, and 10 patients (77%) had no co-morbidities associated with obesity. Six patients (46%) suffered from recurring skin infections on the inner thighs.

Before bariatric surgery, the average BMI was 50 ± 3.5 kg/m^2, and patients gave an average pain evaluation of 7.3 ± 1.4 (VAS range, 5–10; median 7; upper quartile 8.5; lower quartile 6). Within 2 years of the procedure, these patients had lost an average of about 73% of their excess weight (49%–102%), and their BMI had dropped to an average of 32 ± 4.2 kg/m^2. However, most important, the patients reported no improvement in their typical lipedema pain symptoms (VAS 7.9 ± 1.2; VAS range, 6–10; median 8; upper quartile 8.5; lower quartile 7; P = .28;). Lipedema, stage 2 or 3, was diagnosed in all patients after bariatric surgery.

Discussion

Because of a lack of awareness of lipedema, this disease is repeatedly overlooked and misdiagnosed by physicians specializing in other fields [19]. Because lipedema is often associated with obesity, a differential diagnostic assessment (Table 1) is challenging and not part of the routine diagnostics of a bariatric consultation [20,21]. The disease does not display typical edema, as found in lymphedema, and the

Table 1
Differential diagnosis of LiDo and obesity and treatment options

Diagnosis	Assessment	Therapy
LiDo		
	•Family history of LiDo •Only the extremities are affected •Disproportional fat distribution between trunk and extremities •Spontaneous pain, pressure sensitivity, a feeling of tension •Tendency to bruise •Mostly female	•Manual lymphatic drainage •Compression therapy •Skin care •Liposuction
Obesity		
	•Generalized and/or abdominal lipohypertrophy •Male and female	•Nutritional advice •Exercise therapy •Bariatric surgery

LiDo = lipohyperplasia dolorosa.

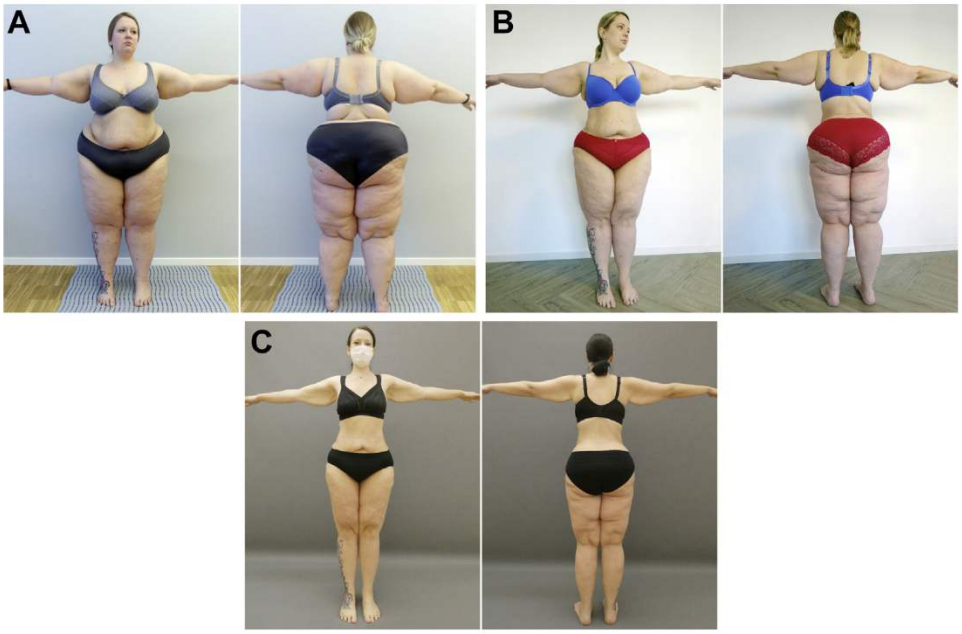

Fig. 3. (A) Before gastric surgery: height, 172 cm; weight, 167 kg; body mass index, 56.4 kg/m^2; LiDo with VAS 5 on arms and legs. (B) Eight months after RYGB: height, 172 cm; weight, 115 kg; body mass index, 39 kg/m^2; WHtR, .5; persistent LiDo with VAS 8 on arms and legs. (C) Twenty months after RYGB: height, 172 cm; weight, 83 kg; body mass index, 28 kg/m^2; WHtR, .47; persistent LiDo with VAS 8 on arms and legs. LiDo with its obligatory pain persists despite percent excess weight loss >50%. The patient requires daily decongestive therapy with manual lymphatic drainage and compression to reduce pain. LiDo = lipohyperplasia dolorosa; VAS = visual analog scale; RYGB = Roux-en-Y gastric bypass; WHtR = waist to height ratio.

traditional term thus is misleading. The neutral descriptive term *lipohyperplasia dolorosa* (LiDo) appears to be more appropriate until its pathophysiology is better understood [16]. Several studies have shown that patients with LiDo have a higher risk of suffering from coincident obesity [5,7,18,22,23], but it is still unclear whether LiDo is an aggravating factor in obesity or the progression of LiDo leads to an increase in BMI. A feature common to both LiDo and obesity is an increase in adipose tissue. In the later stages, patients with LiDo often develop edema and acquire a pronounced disproportionality between the trunk and extremities that is not typical of obesity. However, alimentary obesity of the trunk and extremities gradually eclipses the disproportion, resulting from genetic LiDo, because obesity-related weight gain invariably also leads to large increases in the volume of the affected limbs. This may also

Table 2
Patient characteristics before and after bariatric surgery

Factor	Patient number													Average ± SD
	1	2	3	4	5	6	7	8	9	10	11	12	13	
Lipedema stage	II	II	III	III	III	III	III	III	II	III	III	III	III	
Type of bariatric surgery	RYGB	RYGB	SG	RYGB	RYGB	RYGB	RYGB	RYGB	SG	RYGB	RYGB	RYGB	SG	
Height (m)	1.65	1.72	1.65	1.74	1.68	1.65	1.72	1.68	1.65	1.7	1.79	1.72	1.72	1.7 ± .04
Weight before bariatric surgery (kg)	144	167	150	143	149	139.5	142	139	130	128	149.5	162	144	145.2 ± 10.1
Weight after bariatric surgery (kg)	77	115	110	88	90.5	77	85	69	96	89	110	97	92	92.0 ± 12.7*
BMI before bariatric surgery	53	56	55	47	48	51	48	49	48	44	46	55	49	50.1 ± 3.5
BMI after bariatric surgery	28	39	40	28	32	29	29	24	35	31	34	33	31	31.8 ± 4.2*
%EWL	88	56	49	82	75	87	84	102	55	70	57	74	74	73.3 ± 14.6
%TWL	47	31	27	38	39	45	40	50	26	30	26	40	36	36.7 ± 7.4
VAS before bariatric surgery	6	5	10	8	5	8	7	7	9	9	7	8	6	7.3 ± 1.4
VAS after bariatric surgery	6	8	10	8	8	8	7	7	9	10	8	8	6	7.9 ± 1.2†

RYGB = Roux-en-Y gastric bypass; SG = sleeve gastrectomy; BMI = body mass index; %EWL = percent excess weight loss; %TWL= percent total body weight loss; VAS = visual analog scale (0–10, where 0 = no pain and 10 = very painful).

* $P < .001$ versus before bariatric surgery.

† $P > .05$ versus before bariatric surgery.

have led to the initial overlooking of LiDo in the patients presented here. Studies have shown that typical co-morbidities of obesity, such as arterial hypertension, fat metabolism disorders, and diabetes, are not associated with LiDo [23,24]. Unlike with obesity, patients with LiDo suffer from spontaneous pain, pressure sensitivity, a feeling of tension, and a tendency to bruise in the affected areas, mostly the legs and arms [19,25]. These features should be assessed during bariatric consultation. Determining the BMI alone does not provide sufficient information on the misdistribution of adipose tissue. Particularly where a differential diagnosis is difficult, the waist-to-height ratio, waist-to-hip ratio, and circumference of the extremities should be monitored in addition to the BMI [11,26]. Such monitoring may indicate disproportionality between trunk and extremities, although no cutoff values have been established. Because LiDo is likely to have a genetic basis, the family history of LiDo also should be obtained during bariatric consultation before surgery is considered. The absence of the typical co-morbidities of obesity [23,27] also may indicate a possible LiDo. It is not known how often LiDo is diagnosed only after bariatric surgery and subsequent weight loss. However, where this is the case, because of the decrease in abdominal fat, a slim waist appears, and positive results regarding the improvement in the circumference and volume of the extremities are also reported [28]. Nevertheless, the characteristic cuff or bracelet effect, where fatty tissue abruptly ends at the wrists and ankles, often remains [24].

Here we describe 13 patients with obesity and coincident undiagnosed LiDo. None of the patients were diagnosed with LiDo prior to bariatric surgery, and all continued to suffer from LiDo-associated symptoms after surgery. Importantly, pain, the main feature of LiDo, persisted after bariatric surgery and did not subside. Some similar individual cases have been reported in the literature [24,29,30], but to our knowledge, this has never been studied on a larger scale, especially regarding the persistence of pain.

The urgent question remains of how to treat patients with LiDo and concomitant obesity adequately. The deterioration of mobility due to the additional fatty masses, as well as the increasing hyperinsulinemia due to nonlipedema obesity, worsens the clinical picture of LiDo [27]. Morbid obesity is often the cause of co-morbid secondary lymphedema, and associated changes secondary lymphedema can occur for many reasons, such as after oncologic operations, trauma, and infections, and usually results from a blockage of lymphatic flow. In LiDo, the obesity is coincident and not co-morbid. Furthermore, from hormonal and lymphologic points of view, an anti-inflammatory and weight-reducing diet and exercise are necessarily part of the therapy for both LiDo and obesity [22]. Taken together, we therefore suggest a multidisciplinary approach involving lymphologists, physiotherapists, nutritionists, and surgeons for LiDo and bariatric surgery. Before surgical intervention is considered, patients should receive nutritional counseling and begin complete decongestive therapy, the gold standard of conservative LiDo therapy, that is based on manual lymphatic drainage, compression therapy, and skin care. If these treatments fail, lymphological liposculpture has been shown to be a suitable procedure to reduce the pathologically increased adipose tissue [31].

Three scenarios emerge for the treatment of coincident LiDo and obesity: (1) lymphological liposculpture, that is, removal of the entire diseased tissue on the extremities, which eliminates LiDo; (2) bariatric surgery, which does not change the pain symptoms of LiDo but helps the patient reduce adipose tissue, especially on the trunk; and (3) timed lymphological liposculpture [31] and treatment of obesity (either by diet and exercise or by bariatric surgery), in which case both diseases are cured.

In our daily clinical routine, we recommend LiDo surgery for patients with a BMI of up to 40 kg/m^2, followed by bariatric treatment, if necessary. If the patient's BMI is >40 kg/m^2, we recommend that the obesity be treated first and LiDo afterwards [32] (Fig. 4).

Limitations of the study

The motivation for this work was to draw attention to the problem of the often-overlooked coincident LiDo in patients

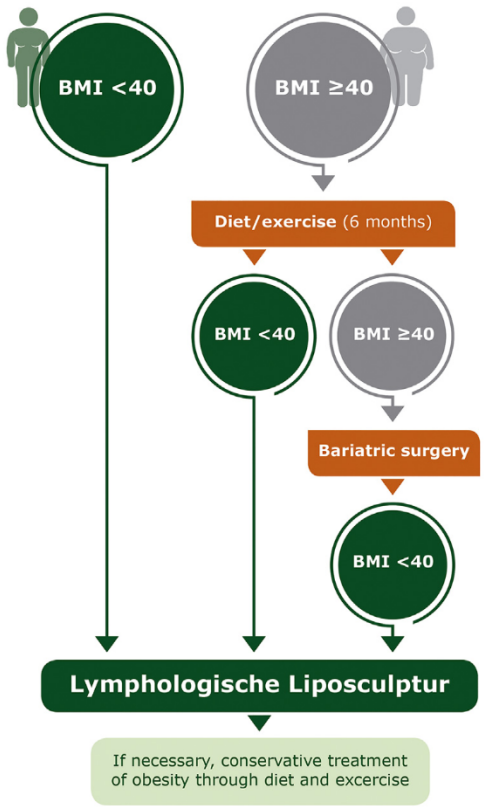

Fig. 4. Treatment scenarios for coincident LiDo and obesity. BMI = body mass index; LiDo = lipohyperplasia dolorosa.

with obesity. However, for this reason, all the patients in this study had been diagnosed with lipedema only after bariatric surgery had taken place. The description of painfulness before bariatric surgery also may be subject to recall bias because we asked patients to recall their symptoms prior to surgery rather than prospectively collecting these data. Furthermore, case numbers are small, and we have data from only 2 treatment centers, which may limit the representativeness of our results. However, because there have been only a total of 3 cases described in the literature to date, none of which considered pain as a symptom, we believe that reporting these cases here is nevertheless of great value. Future studies should investigate weight reduction following LiDo surgery and how often bariatric surgery can be omitted when following this sequence.

Conclusions

LiDo is a painful and debilitating disease. Conservative and operative treatment options are available and can lead to marked improvements in symptoms such as pain, but because of the disease often being concealed by obesity, the onset of relevant treatment can be significantly delayed. Bariatric surgery, used in the treatment of obesity, does not cure lipedema, and as our study shows, even large weight reductions of more than 80% excess weight do not reduce lipedema pain. Following a correct diagnosis, depending on the patient's weight, a treatment sequence focusing on lipedema or obesity first should be chosen.

Disclosure

The authors declare that they have no conflicts of interest.

References

[1] Wold LE, Hines Jr EA, Allen EV. Lipedema of the legs: a syndrome characterized by fat legs and edema. Ann Intern Med 1951;34(5):1243–50.

[2] Wenczl E, Daroczy J. [Lipedema, a barely known disease: diagnosis, associated diseases and therapy]. Orvosi hetilap 2008;149:2121–7.

[3] Forner-Cordero I, Szolnoky G, Forner-Cordero A, Kemeny L. Lipedema: an overview of its clinical manifestations, diagnosis and treatment of the disproportional fatty deposition syndrome—systematic review. Clin Obes 2012;2:86–95.

[4] Buck 2nd DW, Herbst KL. Lipedema: a relatively common disease with extremely common misconceptions. Plast Reconstr Surg Global Open 2016;4(9):e1043.

[5] Child AH, Gordon KD, Sharpe P, et al. Lipedema: an inherited condition. Am J Med Genet A 2010;152A:970–6.

[6] Földi E, Földi M. Lipedema. In: Földi E, Földi M, editors. Foldi's textbook of lymphology. 2nd ed., Munich: Elsevier; 2006. p. 417–27.

[7] Marshall M, Schwahn-Schreiber C. Prevalence of lipedema in working women in Germany (Lipedema 3 Study). Phlebologie 2011;3:127–34.

[8] Gensior MH, Cornely M. [Pain in lipoedema, fat in lipoedema and its consequences: results of a patient survey based on a pain questionnaire]. Handchir Microchir Plast Chir 2019;51(4):249–54.

[9] Cornely M, Gensior M. Lipedema update 2014: Cologne Lipodema Study. LymphForsch 2014;18:66–71.

[10] Al-Ghadban S, Cromer W, Allen M, et al. Dilated blood and lymphatic microvessels, angiogenesis, increased macrophages, and adipocyte hypertrophy in lipedema thigh skin and fat tissue. J Obes 2019;2019:8747461.

[11] Reich-Schupke S, Altmeyer P, Stucker M. Thick legs: not always lipedema. J Dtsch Dermatol Ges 2013;11:225–33.

[12] Rapprich S, Loehnert M, Hagedorn M. Therapy of lipoedema syndrome by liposuction under tumescent local anaesthesia. Ann Dermatol Venereol 2002;129:1S711.

[13] Reich-Schupke S. Die besondere rolle der adipositas in der lymphologie. Vasomedicine 2014;5:230–6.

[14] Okhovat JP, Alavi A. Lipedema: a review of the literature. Int J Lower Extremity Wounds 2015;14:262–7.

[15] Brenner E. Lipödem: lipohyperplasia dolorosa. Phlebologie 2005;34.

[16] Cornely M. Terminologie des Lipödems. Phlebologie 2005;35:271.

[17] Kayserling E. *On the histology of lipedema. In: Strößenreuther RHK, editor. Lipedema and cellulitis and other diseases of the adipose tissue*. Cologne: Viavital Verlag; 2001.

[18] Wollina U, Heinig B. Treatment of lipedema by low-volume microcannular liposuction in tumescent anesthesia: results in 111 patients. Dermatol Ther 2019;32:e12820.

[19] Cornely M. Fatter through lipids or water: lipohyperplasia dolorosa versus lymphedema. Am J Cosmet Surg 2014;31:189–95.

[20] Reich-Schupke S, Schmeller W, Brauer WJ, et al. S1 guideline lipedema. J Dtsch Dermatol Ges 2017;15(7):758–68.

[21] Rapprich S, Dingler A, Podda M. Liposuction is an effective treatment for lipedema: results of a study with 25 patients. J Dtsch Dermatol Ges 2011;9:33–40.

[22] Faerber G. Nutritional therapy in lipedema and obesity - results. Of a guideline-based therapeutic concept. Vasomed 2017;29:176–7.

[23] Sanchez-De la Torre Y Torre YS, Wadeea R, Rosas V, Herbst KL. Lipedema: friend and foe. Horm Mol Biol Clin Investig 2018;33(1):1–10.

[24] Georgiou I, Kruppa P, Schmidt J, Ghods M. Liposuction for lipedema: functional therapy or aesthetic procedure? Aesthetic Plast Surg 2021;45(1):212–3.

[25] Cornely M. Lipedema and lymphedema. In: Plewig G, Prinz J, editors. *Progress in practical dermatology and venereology*. Berlin: Springer; 2003. p. 255–63.

[26] Damstra R, Habbema L, Hendrickx A. Lipedema: guidelines in the Netherlands 2014. In: Damstra R, editor. Diagnostic and therapeutic aspects of lymphoedema 2013, Bonn: Rabe Verlag; 2014.

[27] Faerber G. Obesity and chronic inflammation in phlebological and lymphatic diseases. Phlebologie 2018;47:55–65.

[28] Fink JM, Schreiner L, Marjanovic G, et al. Leg volume in patients with lipoedema following bariatric surgery. Visc Med 2021;37(3):206–11.

[29] Bast JH, Ahmed L, Engdahl R. Lipedema in patients after bariatric surgery. Surg Obes Relat Dis 2016;12:1131–2.

[30] Pouwels S, Huisman S, Smelt HJM, Said M, Smulders JF. Lipoedema in patients after bariatric surgery: report of two cases and review of literature. Clin Obes 2018;8:147–50.

[31] Cornely M. Lymphological liposculpture. Der Hautarzt 2007;58:653–8.

[32] Association of Scientific Medical Societies. S3 guideline: surgery for obesity and metabolic diseases. Version 23, Springer; 2018.