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Case Report

Lipoedema in patients after bariatric surgery: report of two cases and review of literature

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Summary

Lipoedema is a disorder of adipose tissue that is characterized by abnormal subcutaneous fat deposition, leading to swelling and enlargement of the lower limbs as well as the trunk. This entity is often misdiagnosed as lymphoedema or obesity and, therefore, may be overlooked and missed in patients scheduled for bariatric surgery. Patients with lipoedema who undergo bariatric surgery may have to continue to have extensive lower extremity and trunk adiposity despite adequate weight loss. In this report, we present two patients who had extensive trunk and lower extremity adiposity, one of them before and the other after the bariatric surgery.

Keywords: Bariatric surgery, differential diagnosis, lipoedema, obesity.

Introduction

Lipoedema is a disorder of adipose tissue that is characterized by abnormal subcutaneous fat deposition, leading to swelling and enlargement of the lower limbs and also the trunk (1, 2). Lipoedema is a distinct entity with possible inheritance of X-linked dominant or autosomal dominant pattern with sex limitation (1–3). This condition is often misdiagnosed as lymphoedema or obesity and, therefore, may be overlooked and missed in patients scheduled for bariatric surgery. Lipoedema has a mean prevalence of 11% among women in the Western world (4).

Lipoedema is a soft, minimally pitting entity that lacks epidermal changes, extending symmetrically from the buttocks or lower extremities to the ankles (1, 5, 6). Important aspect of lipoedema is that it spares the feet (1, 3, 5, 7, 8). It is very important to distinguish lipoedema from obesity and vice versa. This is because of the different treatment modalities for both entities, and secondly patients with lipoedema scheduled for bariatric surgery may have different results in terms of weight loss. Additionally, after bariatric surgery the disproportionate adiposity might be attenuated.

Patients with lipoedema who undergo bariatric surgery may have to continue to have extensive lower extremity and trunk adiposity despite adequate weight loss. In this report, we present two patients who had extensive trunk and lower extremity adiposity, one of them after and one before the bariatric surgery. Both of the conditions were refractory to weight loss after bariatric surgery. Secondly, we would like to elucidate that a combination of lipoedema and obesity provides challenges in patients admitted to bariatric surgery. Informed consent was obtained from all the individual participants included in the study.

Case 1

A 35-year-old female was admitted to the bariatric surgery clinic (Fig. 1a). She had a height of 166 cm and a weight of 159 kg, resulting in a body mass index (BMI) of 57.5 kg/m^2. Prior to surgery, the circumference of her upper legs were 78 and 83 cm, and both her calves were 54 and 55 cm. Both of them were tender to palpation. There were no obesity-related comorbidities, no family

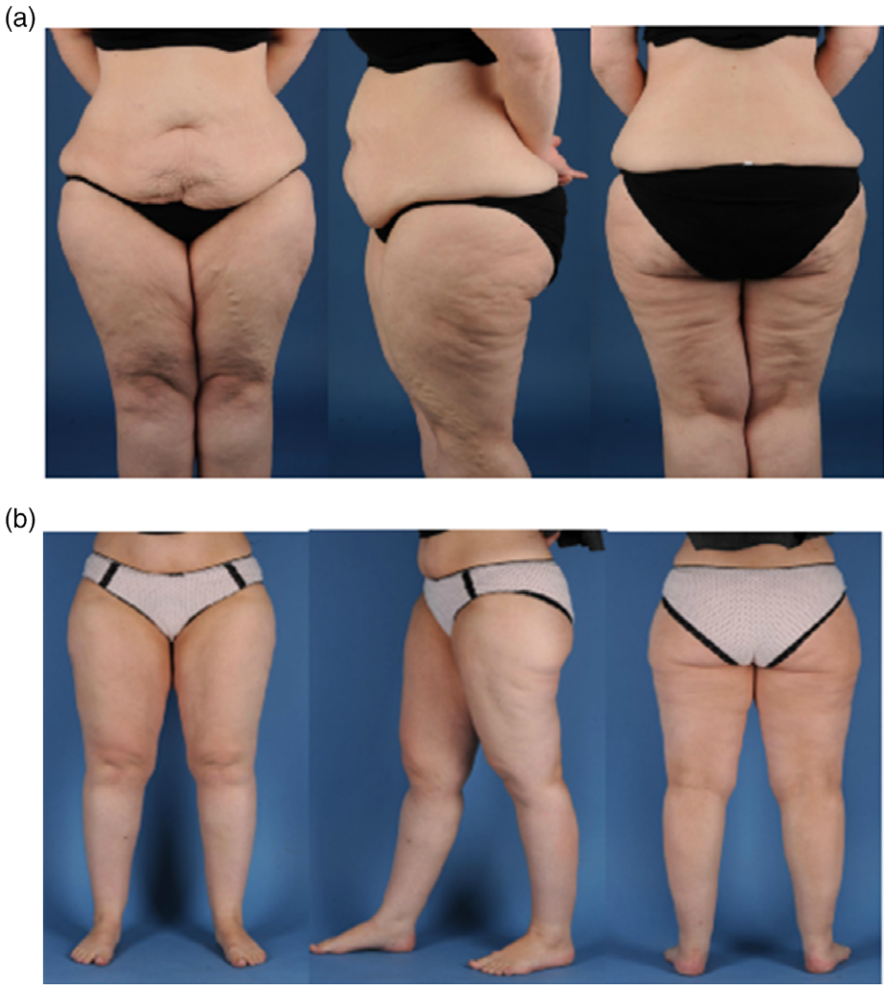

Figure 1 Pictures of the body composition. (a) After the sleeve gastrectomy (Case 1). (b) After the Roux-en-Y gastric bypass (Case 2).

history of any lymphatic disease and no previous skin infections. The patient had undergone a sleeve gastrectomy. After her initial surgery, she lost 64 kg, resulting in a BMI of 34.2 kg/m^2. The circumference of her upper legs were 82 and 82 cm, and both her calves were 54 and 54 cm. In the first 2 years after her sleeve gastrectomy, she underwent plastic surgery in terms of a lower body lift and an upper thoracic lift with breast reconstruction (Rubin's technique). In the years following, she had a weight regain of 22 kg (BMI 34.2 kg/m^2) to 117 kg (BMI 42.5 kg/m^2). The circumference of her upper legs (82 and 82 cm) and both her calves (54 and 54 cm) did not increase during this time period. Due to this weight regain, she had revisional bariatric surgery, approximately 5 years after her sleeve gastrectomy. The sleeve gastrectomy was converted to a single anastomosis duodeno-ileostomy. Three months after the surgery, she had a BMI of 38.8 kg/m^2 (106.9 kg) and no decrease in circumference of her upper legs and calves. Long-term treatment was carried out with compression stockings.

Case 2

A 24-year-old female was admitted to the bariatric surgery clinic (Fig. 1b). She had a height of 163 cm and a weight of 108 kg, resulting in a BMI of 40.6 kg/m^2. There were no obesity-related comorbidities (no type 2 diabetes mellitus) and no history of skin infections. She had history of lymphoedema that was resistant to therapy by a certified physiotherapist. In the next 18 months, she was diagnosed with lipoedema and had six liposuction surgeries with moderate results. These liposuction surgeries were performed by an experienced plastic surgeon in other Dutch hospital. However, hospital records were obtained and she had underwent six times liposuction (technique as proposed by Stutz and Krahl (10) and one time lipectomy by an experienced plastic surgeon).

Despite some weight loss, she had a Roux-en-Y gastric bypass 3 years after her initial lipoedema diagnosis. Four months after the surgery, she lost 73.9 kg, resulting in a BMI of 27.8 kg/m^2. Unfortunately, 6 months after the

Table 1 Clinical differences between obesity, lipoedema and lymphoedema (derived from Stutz and Krahl *et al.* (10)).

	Obesity	Lipoedema	Lymphoedema
Patient history			
Sex	Male and female	Female	Male and female
Family history positive	Common	Common	Present in primary lymphoedema
Progression	All over the body (mostly limited to the trunk)	Involved areas	Proximally, from distal portion of the limb
Physical examination			
Bilateral involvement	Always	Always	Primary: often Secondary: rare
Foot involvement	Common	Absent	Common
Malleolar fat pad	Absent	Present	Absent
Consistency on palpation	Soft	Soft to firm	Firm
Pitting oedema	Absent	Minimal	Always present in variable severity
Pain on pressure	Absent	Common	Absent

surgery the circumference of her upper legs increased from 66 to 76 cm (right side) and from 61 to 71 cm (left side). Also the circumference of her calves increased from 43 to 48 cm (right side) and from 42 to 47 cm (left side). Both of them were tender to palpation. She used compression stockings for both legs as long-term treatment.

Discussion

Here we report two challenging cases of obesity and lipoedema: one case that developed lipoedema after bariatric surgery and one case with a history of lipoedema.

First described in 1940 by Wold *et al.* (8), little is understood about lipoedema. It is described as 'adiposis dolorosa' (in other word, painful fat) and is related to a more extreme adipose tissue disorder, Dercum's disease (8, 9). Mainly women are affected by lipoedema and the incidence in the United States is approximately one in nine adult women, resulting in millions of women affected by this disease (1, 9). Unfortunately, little is known about the aetiology of lipoedema and it is frequently not recognized by physicians or even misdiagnosed as obesity or lymphoedema (1, 9). Mainly physicians do not even know what kind of advice they need to give to these patients, which causes desperation for both the patient and the physician). Far too often, the women who suffer from lipoedema are told that their leg growth and swelling is a result of their inability to control their diet or of their sedentary lifestyles. As if to suggest acceptance, they are often told that the women in their families just 'have big legs'. When the disease progresses, they are frequently questioned about their compliance with diet and exercise recommendations (9). This can lead to severe psychosocial distress, including anxiety, depression, eating disorders and isolation (9). One common misconception is that these patients suffer from lifestyle- or diet-induced obesity (8, 9). Although lipoedema can co-exist next to obesity in some patients and the obesity may influence subcutaneous adipose tissue, lipoedema needs to be considered as a separate entity/diagnosis. Unlike obesity, the adipocyte hypertrophy and swelling associated with lipoedema are resistant to change with diet and exercise or bariatric surgery and caloric restriction (1–3, 9).

In clinical practice, it is difficult to determine whether a patient had lipoedema, lymphoedema or obesity. Lipoedema is a soft, minimally pitting entity that lacks epidermal changes. Also it extends symmetrically from the buttocks or lower extremities to the ankles (1, 5, 6). Important aspects of lipoedema are that it spares the feet and it has a normal lymphoscintigraphy (1, 3, 5, 7, 8).

Table 1 gives an overview of the clinical differences between obesity, lipoedema and lymphoedema. As the lipoedema progresses, it becomes easier to recognize. The diagnostic criteria for lipoedema were first described by Wold in 1951 and have been modified in the recent years by Buck and Herbst (8, 9). The disease affects mainly women in the third decade of life. Characteristically, the feet are spared and the fat deposits begin abruptly above the malleoli, causing a sharp demarcation between normal and abnormal tissue at the ankle ('cuff sign'). A typical explanation by patients is a body with disproportionate halves. Subcutaneous adipose tissue of patients with lipoedema is non-pitting and stops at the ankle; it is granular to palpation; has a 'grain-like' or 'nodular (beans in a bag)' structure and the adipose tissue is tender to palpation and pressure (8, 9). Skin is easily bruised and may have telangiectasia due to fragile subdermal capillaries (9).

Lipoedema can be divided into five types of which types 1–3 are the most common (9). Also patients can have a mixture of several types. Type 1 is situated at pelvis, buttocks and hips; type 2 from buttocks to knees with the formation of fold of fat at the inner side of the knee; type 3 occurs from buttocks to ankles; type 4 is present only at the arm and finally, type 5 is isolated to the lower legs (8, 9).

Lipoedema has a tendency to progress over time such that disease severity can be described in stages. There are currently four reported stages of lipoedema: (i) involves an even skin surface with an enlarged hypodermis; (ii) involves an uneven skin pattern with the development of a nodular or mass-like appearance of subcutaneous fat, lipomas and/or angiolipomas; (iii) involves large growths of nodular fat, causing severe contour deformity of the thighs and around the knee and (iv) involves the presence of lipolymphoedema (1, 3, 4, 9).

In terms of aetiology, we do not completely understand the occurrence of lipoedema. Looking at histological studies, it is proposed that the initial swelling in lipoedema is the result of two factors: adipocyte hypertrophy and hyperplasia (9). Secondly there is thickening of the interstitium with increased interstitial fluid, due to the elevated hydrostatic pressure. Although there is an increase in interstitial fluid in the early stages of the disease, it does not compromise the function of the lymphatic system (8, 9). Therefore, it is rather likely that the 'oedema' is secondary to the overwhelming lymphatic pump, rather than a true dysfunction (9). When the disease progresses, it is suggested that lymphatic channels tend to stretch and dilate, leading to the development of 'micro-aneurysms', which have a tendency to leak. Finally, in addition to adipocyte hypertrophy, thickening of the interstitium and lymphatic changes, the subdermal vascular plexus undergoes changes consistent with microangiopathy. This leads to fragile capillaries that correspond to the easy bruising and telangiectasia seen in lipoedema patients (6, 9).

The aim of treatment is to optimize patient mobility, reduce complaints and improve overall quality of life. The importance of weight control is emphasized in all patients. Conservative therapy, such as manual lymphatic drainage and orthopaedic or psychological counselling, is started on indication. Treatment of lipoedema often includes compression stockings or liposuction, which is at this moment the only available technique to correct the abnormal adipose tissue (6, 8, 9).

Before bariatric surgery, the difference between lipoedema and obesity can be challenging, especially in female patients with disproportionate lower trunk or lower extremity subcutaneous fat. Identification of patients with lipoedema before bariatric surgery may help in guiding expectations of weight loss after surgery. If suspected, a lymphoscintigraphy can be conducted as an aid to distinguish between lipoedema, lymphoedema and obesity (1). However, due to the rare nature of lipoedema in our bariatric practice, these investigations are conducted rarely. It is important for clinicians working in bariatric medicine to pay attention to patients with a very disproportionate subcutaneous fat distribution. As it is very unlikely that significant excess weight loss will resolve trunk and extremity fat alone, additional procedures might be needed to remove this excess, such as liposuction. Interestingly, the first case showed no difference in circumference of the upper legs and the calves after surgery, and the second case showed an increase of circumference of the upper legs and calves. A possible explanation can be that the regular liposuction surgeries damaged the lymphatic tissue, resulting in an increased circumference (1, 10). Despite the fact that the procedures were performed by an experienced surgeon, it still need to be that liposuction and lipectomy techniques need to be performed by an experienced personnel.

Conclusion

In obese patients scheduled for bariatric surgery, the presence of lipoedema can be challenging. If there is a combination of obesity and lipoedema present in a patient prior to bariatric surgery, or a suspicion of lipoedema, patients need to be elucidated about the fact that we need to strive for comorbidity control and increasing quality of life. This is despite the fact that some of the patients have a desire for a cosmetically more attractive result.

Conflict of Interest Statement

No conflict of interest was declared.

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