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Case Report

Lipedema subsequent with lymphedema and obesity in male patient: a rare case

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ABSTRACT

Lipedema is a disorder of abnormal subcutaneous fat deposition which almost exclusively occured in women. It is an unusual case that happens in male patients. Lipedema is often misdiagnosed to obesity and lymphedema. In case at hand, a 39-year-old obese male (body mass index of 54.3 kg/m^2) presented bilateral non-pitting edema from buttocks to knees, palpable fat tissue nodules, pain at pressure, positive cuff sign, negative Stemmer-Kaposi sign, hyperpigmentation and thickened skin along the calves, and minimal responds to compression therapy. Average blood pressure was 145/90 mmHg. Laboratory studies were unremarkable, except leukocytosis and increased erythrocyte sedimentation rate which might be related to obesity-associated leukocytosis. Diagnosis of type III lipedema subsequent with lymphedema, grade III obesity, and mild hypertension was made. The patient subsequently reported an improvement of the complaints after receiving nonsteroidal anti-inflammatory drug, proton pump inhibitors, and antihypertensive. Weight loss was encouraged as initial steps to reduce aggravating risks of obesity. This case underlines that lipedema needs to be considered as a differential diagnosis in male patients.

Keywords: Lipedema, Lymphedema, Obesity

INTRODUCTION

Lipedema is a chronic disease which is characterized by enlargement of extremities, mainly in lower limbs, due to abnormal fat deposition. Fat deposition commonly extends from shoulder to wrist and from hip to ankle, sparing the hands and feet, which could be a clinical manifestation to differentiate from obesity and lymphedema. Lipedema almost exclusively occured in women. Men with lipedema have been brought up only in case reports. Pathogenesis and etiology of lipedema is still poorly understood. Diagnosis of lipedema mainly relies on history taking and physical examination. On other sides, lipedema is often underdiagnosed by healthcare professionals and often misdiagnosed to obesity and lymphedema. Considering the enormous differential diagnosis of enlargement of bilateral lower limbs, a mindful investigation should be done. Treatment is often challenging. However, conservative treatment using weight reduction and compression bandaging is effective in early stages and surgical interventions by liposuction is effective in advanced stages.[1,2] In the report at hand, we report a rare presentation of lipedema in a male patient.

CASE REPORT

A 39-year-old male presented with painful legs since two weeks ago. Pain predominantly felt in soles of both feet, and also felt in both calves. Pain was aggravated by standing for prolonged periods of time. There is also symmetrical enlargement of both legs that began above the ankles and extended proximally to the upper thighs. The symptoms had developed two years ago as he subsequently began to steadily gain weight. Two weeks ago, both swollen legs had a dimple (or pit) after being pressed for a few seconds. Treatment with bed rest and compression stocking was a little help in reducing the swelling in the leg and the pressed-pit, yet the pain was still intense. The patient's past medical, medication, and

family history were unremarkable. He was admitted for further investigations.

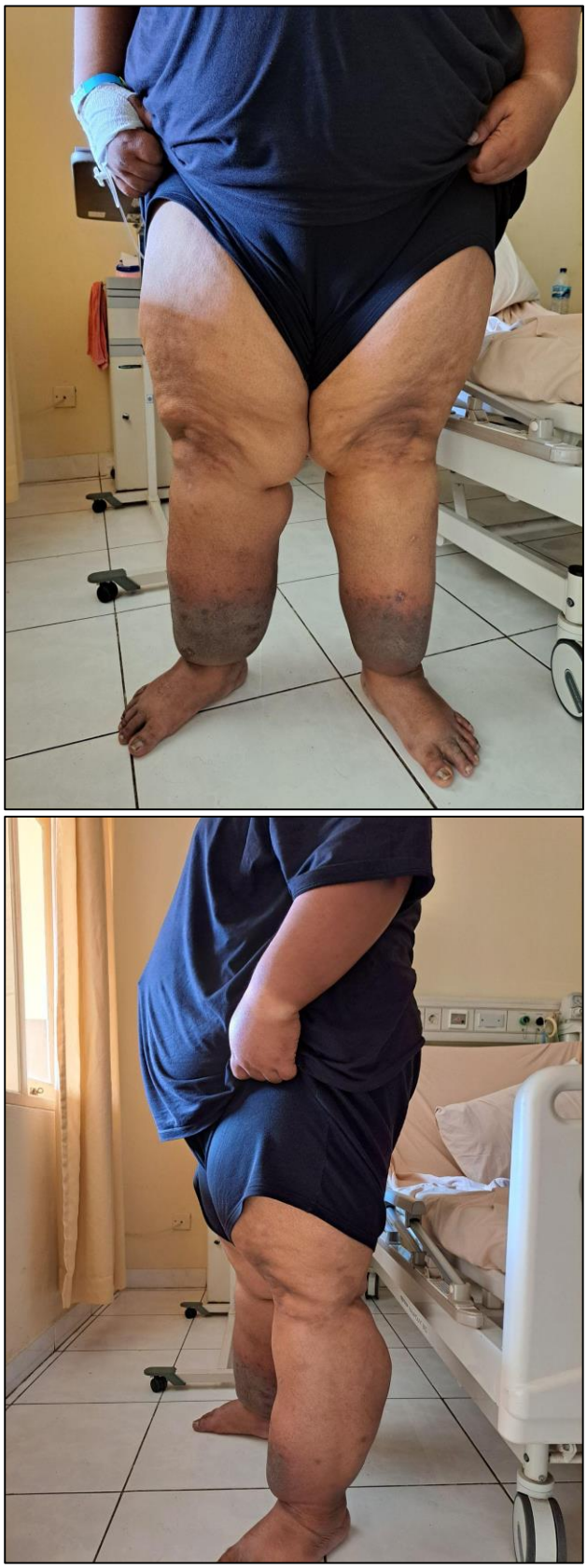

Figure 1: Abnormal fat deposition from buttocks to ankles. Note the ankle pad or cuff sign.

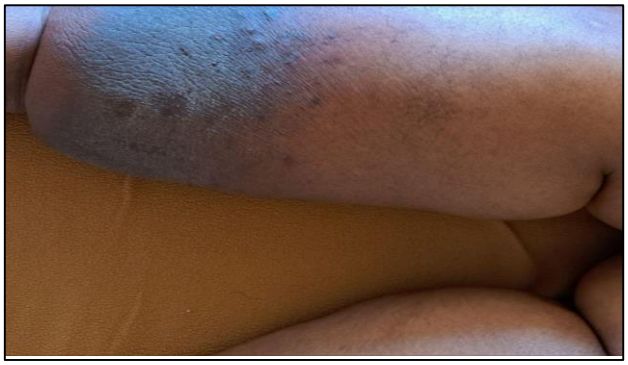

Figure 2: Thickened skin, hyperpigmentation, and warty nodules seen along the calves.

Bloodwork for hematology revealed WBC of 18.32×10^3 µL (normal range 4.0-10.00) with neutrophils count dominance and erythrocyte sedimentation rate (ESR) of 88 mm/hours (normal range 0-20). Other laboratory studies, including biochemistry (liver function test, renal function test, uric acid, lipid profile), clotting studies, serum electrolytes, thyroid function tests were normal. A chest radiograph revealed normal lungs and heart. Femur, cruris, and pedis radiograph revealed soft tissue swelling. Sonography of the abdomen revealed parenchymal liver disease, cholelitiasis, and splenomegaly.

Diagnosis of type III lipedema with subsequent lymphedema, grade III obesity, and mild hypertension was made. During hospitalization, the medication of NSAID (mefenamic acid 500 mg every 8 hours), proton pump inhibitors (omeprazole 40 mg every 12 hours), and antihypertensive (candesartan 8 mg every 24 hours) were given. The patient subsequently reported an improvement of the complaints. After explaining diagnosis and treatment options, the patient was initially advised to do conservative treatment using a weight reduction program and compression bandaging.

DISCUSSION

Diagnosis of lipedema is commonly made on clinical reasoning after exclusion of other differential diagnoses. Common causes of bilateral pitting edema of lower limbs include congestive heart failure, chronic kidney disease, cirrhosis, and hypoalbuminemia. Clues which lead to diagnoses above can be present in history-taking and physical examination, for example, jugular vein distention, dyspnea, pleural effusion, ascites. Bilateral non-pitting edema could be caused by thyroid hormone imbalance, an unusual manifestation of Grave's disease. Medication-induced edema caused by calcium channel blocker (e.g amlodipine), gabapentin, non-steroidal anti-inflammatory drugs (e.g ibuprofen), oral contraceptives, corticosteroids (e.g prednisone), thiazolidinediones.[2] History-taking, physical examination, and laboratory studies of differential diagnosis above were all negative in this patient. We conclude the possibility of diagnosis of the patient were lipedema, lymphedema, and obesity.

Lipedema is frequently misdiagnosed as lymphedema or obesity.[2] Despite similar phonetic sounds of both diagnoses, lipedema has distinct entities compared to lymphedema. Moreover, lipedema might proceed to involve lymphatic and venous systems, which lead to confusion in the diagnosis.

Lipedema is a disorder of abnormal subcutaneous fat deposition which exclusively occured in women. Clinical criteria for the diagnosis of lipedema, are (1) chronic, bilateral, symmetrical, disproportionate fatty tissue hypertrophy on the limbs, minimal involvement of hands and feet (cuff sign), (2) pain on pressure and touch, or after standing or sitting after prolonged period, (3) mild pitting edema that can improve with elevation, (4) marked tendency to develop spontaneous hematomas, (6) persistent enlargement after elevation of the extremities or weight loss, (7) telangiectasia and visible vascular markings around fat deposits, (8) "negative" Stemmer-Kaposi sign (whereas "positive" Stemmer-Kaposi sign could found in later stage of lymphedema).[1,3] Based on the distribution of fat, lipedema is classified into five types; type I: pelvis, buttocks, and hips; type II: buttock to knees; type III: buttocks to ankles; type IV: arms; type V: knees to ankles. Furthermore, lipedema is classified into four stages by its severity; Stage 1: the skin surface is normal, and the subcutaneous fatty tissue has a soft consistency but multiple small nodules can be palpated; stage 2: the skin surface becomes uneven and harder due to the increasing nodular structure (big nodules) of the subcutaneous fatty tissue (liposclerosis); stage 3: hardening and thickening of the subcutaneous with lobular deformation; stage 4: lipedema with lymphedema, called lipolymphedema.[1,4,5]

Lymphedema is accumulation of protein-rich fluid in interstitial tissue due to lymph drainage failure. On the contrary of lipedema, lymphedema may be asymmetrical, unaccompanied by easy bruising or pain or tenderness. Hands and feet are frequently affected. Pitting edema is present in early stages, then diminishes at later stages. Involvement of skin inflammation, warty changes, fibrosis, dermal thickening or induration are common. Stemmer-Kaposi sign, or the inability to pinch the base of the second toe, is a distinctive characteristic for later stages of lymphedema. Lymphedema generally responds to compression therapy, while lipedema poorly responds to compression therapy.[6-8]

Obesity has a symmetrical distribution of fat with the involvement of the trunk and common involvement of arms. "Cuff sign" is negative in obesity. Hematoma tendency and tenderness or pain are absent. Obesity is influenced by diet, while in contrast lipedema is not. Obesity does not respond to compression therapy.[2,9]

In case at hand, an obese male (BMI of 54.3 kg/m^2) presented bilateral non-pitting edema from buttocks to knees, palpable fat tissue nodules, pain at pressure, positive cuff sign, negative Stemmer-Kaposi sign, hyperpigmentation and thickened skin along the calves, and minimal responds to compression therapy. Diagnosis of type III lipedema subsequent with lymphedema and grade III obesity was made.

Laboratory studies show leukocytosis with neutrophilia and increase in ESR. No other causes for leukocytosis (e.g. infection, inflammation, trauma, smoking, drugs, malignancy, pregnancy, etc.) was identified. This finding might be related to obesity associated-leukocytosis. There are sufficient studies which prove adipose tissue is a source of inflammatory mediators. Pro-inflammatory states of obesity (elevated ESR) actively contribute to alterations in hematologic markers, i.e. reactive neutrophilia.[10,11]

Treatment of lipedema is difficult and challenging. Lipedema has negative impacts on self-esteem, mobility, and quality of life. Priority of treatment in lipedema is improvement of signs and symptoms, to stop or slow progression of the disease, and prevent complications. Lipedema management needs an integrative and multidisciplinary approach, including weight control, physical activity, dietary counseling, improving mobility, pain management, and psychosocial support related to lipedema. Obesity is an aggravating factor, as the prevalence of lipedema increases progressively with weight gain. Even though weight loss exhibits a minimal effect on lipedema volume, it has shown more benefits: reduced symptoms, better mobility, and better general health status. Moreover, potentially irreversible damage can arise in the lymphatic system due to obesity, particularly if BMI exceeds 50, leading to overload and the formation of lymphedema. Prophylactic weight loss is a necessity to avoid the aggravation of the condition. For patients with minimal to no benefits following conservative approach, surgical intervention is an option. Surgical options which could be considered are liposuction and lipectomy. These methods can remove existing fat deposit, but further deposit must be prevented.[4,12]

In the present case, treatment is emphasized to alleviate pain and control blood pressure. Medication of NSAID, proton pump inhibitors, and antihypertensive were given. Compression bandaging and leg elevation was suggested to reduce lymphedema. After a few days of hospitalization and bed rest, pain was diminished and leg swelling was reduced. Weight loss was encouraged as initial steps to reduce aggravating risks of obesity.[13] Continual follow-up must be done to maintain quality of life and prevent disease progression.

CONCLUSION

We reported a rare case of lipedema subsequent with lymphedema and obesity in a male patient. This case underlines that lipedema needs to be considered as a differential diagnosis in male patients.

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