

MASSIVE LOCALIZED LIPOLYMPHEDEMA PSEUDOTUMOR IN A MORBIDLY OBESE PATIENT

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ABSTRACT

We describe a 31 year old man with a massive localized tumor-like lipolymphedema, a puzzling entity that afflicts the morbidly obese. The 281 kg man presented with a growing ulcerated bleeding mass located on his proximal medial thigh and suspicious for sarcoma. After en bloc resection of the 28.2 kg edematous mass, no evidence of neoplasm was found, only prominent lymphatic vessel dilation and edema with large quantities of unremarkable adipose and connective tissue. The lesion conformed to the diagnostic criteria for massive localized lipolymphedema (MLL) pseudotumor.

Keywords: lipolymphedema, obesity, adipose tissue, lymphedema, lipedema, liposarcoma

Massive localized tumor-like “lipolymphedema,” a puzzling entity afflicting predominantly morbidly obese individuals, has been described by Földi et al (1) and alternatively termed “massive localized lymphedema in the morbidly obese” by Farshid and Weiss (2). Additional cases have been described by Wu et al (3) with a cautionary note about misdiagnosis, i.e., confusion with well-differentiated or low-grade liposarcoma. Clinical and pathologic features together substantiate the diagnosis. This report concerns an unusually large localized area of lipolymphedema presenting acutely as an expanding medial proximal

thigh “tumor” in an obese man. The mass was resected successfully en bloc without recurrence.

CASE REPORT

A 31 year old depressed, obese man presented with a bleeding right medial thigh mass. The painless mass had been increasing in size for 3 years and developed an ulcer that started to weep blood-tinged fluid one day prior to presentation. He denied any fever or chills. The patient weighed 281 kg with a height of 1.75 meters. Mobility had been severely limited because of the mass as well as his body weight. Past medical history was significant for long standing obesity, a requirement of 8L of oxygen via nasal continuous positive airway pressure at home, and nocturnal bilevel positive airway pressure for obstructive sleep apnea. No prior operations or irradiation were reported, there was no significant family history, and the patient was taking no medications. He had been smoking one pack of cigarettes a day for 20 years but denied alcohol use. Physical examination revealed a large mass extending from the right medial groin to the middle medial posterior thigh (*Fig. 1*). The posterior aspect of the mass displayed a 10 cm ulcer with a central necrotic eschar and surrounding erythema. The skin overlying the mass had a peau d’ orange appearance with nodules of woody texture posteriorly. He was afebrile with a normal white blood cell count and



Fig. 1. Medial thigh mass with thickened skin and woody changes. Ulcer not seen in this image due the enormity of the mass.



Fig. 2. Mass excised en bloc and lifted with Steinman retention pins from ceiling.

routine laboratory screen. No special imaging studies were performed because of the patient's extreme weight and size. The pre-operative diagnosis was possible liposarcoma.

Surgical resection was undertaken with two surgical teams on a semi-urgent basis because of hemorrhage from the ulcerated mass and to circumvent necrotizing infection. Once the patient was anesthetized, the right inner thigh mass was suspended from the ceiling with Steinmann pins (*Fig. 2*). One

team began excising the mass from the lateral to medial aspect; the second team excised from the medial aspect. During the resection, there were multiple large feeding blood vessels encountered, which were ligated with ties and clips (*Fig. 3*). The soft tissue was described as edematous with gross visualization of "prominent lymphatic channels" requiring ligation. The two dissection planes were then connected and the mass removed en bloc. The patient required 8200 ml of

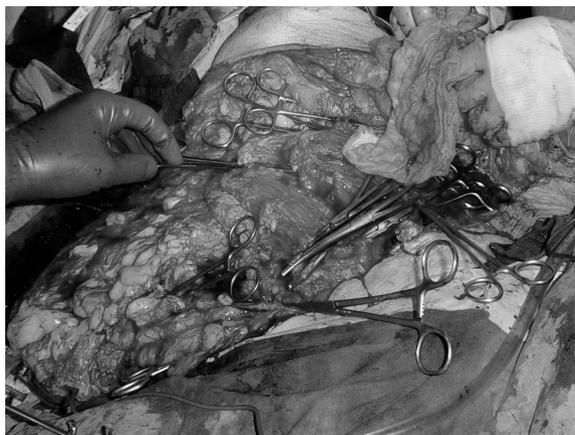


Fig. 3. Ligation of large and prominent blood and lymphatic vessels.



Fig. 4. Mass completely excised. Wound closed primarily and JP drains left in place.

crystalloid with an estimated blood loss of 300 ml. The mass measured 68 x 63 x 9 cm and weighed 28.2 kg (after considerable loss of blood and draining lymph). Three Jackson-Pratt (JP) drains were placed and the wound closed in layers (*Fig. 4*). The patient emerged from anesthesia and was monitored in the Intensive Care Unit postoperatively, then transferred to the ward, where he was treated with a 10 day course of Ciprofloxacin for a wound infection. The proximal wound was opened, and Dakin's solution was used to dress the wound three times a day. Prior to discharge home, two JP drains were discontinued, his wound was granulating well without erythema, and he

was able to get out of bed with the assistance of a walker. The follow-up clinic visit was significant for a smiling, ambulating patient, whose wound was healing well. He was continuing to lose weight.

Histologic examination (*Fig. 5*) displayed prominent lymphatic vessel dilatation in the skin and subcutaneous tissue, multifocal folliculitis and follicular abscesses, and an inflamed ulcer bed with acute inflammation and granulation tissue. There was no evidence for neoplasm. During sectioning, "markedly edematous tissue weeping clear fluid" was noted. The deeper layers contained large quantities of unremarkable adipose and connective tissue.

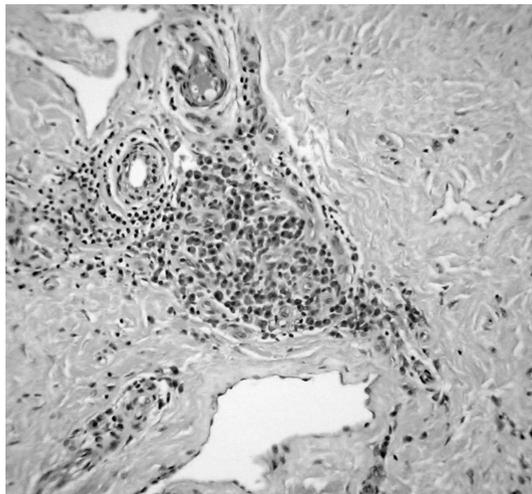


Fig. 5. Subcutaneous tissue containing several dilated lymphatic vessels with nearby vein and artery (20X). Note endothelial cells lining dilated lymphatic vessels and acute inflammatory response.

DISCUSSION

Massive localized lymphedema (MLL) in the morbidly obese has been termed “pseudosarcoma of the morbidly obese” by Goshtasby et al (4) and “localized and tumor-like lipolymphedema” by Földi et al (1). This unusual entity afflicts mostly morbidly obese individuals, affecting both females and males with a female predominance (1). Locations include the abdominal wall, and usually unilateral involvement of the thighs, inguinal region, knees, scrotum, arms and one reported instance in the foot (5). History of precipitating trauma such as lymphadenectomy, vein stripping or other operations may be absent. Treatment is often sought when the lesion has reached a size that inhibits daily activity and is associated with wound ulceration or infection.

It is important to distinguish MLL from hernias and lipomatous tumors, especially liposarcoma, given its large and expanding size. Both Farshid et al (2) and Wu et al (3) suggest that one of the distinctive histologic features is “reactive vessels at the border of the adipose and fibrous tissue.” This observation suggests a localized lymphatic

obstruction where lymphatic fluid accumulates, stimulating tissue growth factor release and eventual appearance of hypertrophic chronic lymphedema.

Malignancy must be ruled out, and prompt resection is warranted in suspicious lesions. In the cases reported by Farshid et al (2), Wu et al (3), and Barr et al (6), surgical resection was performed for recurrent infection or severe limitation of daily activities. Surgical excision has been recommended as the treatment of choice for MLL particularly if the lesion is localized and very large, or to rule out possible malignancy or prevent necrotizing infection. In Wu’s series, 6 of 12 patients developed a local recurrence within 10 months to 10 years after excision (3).

Thus, MLL is a benign lesion of unclear pathogenesis that is potentially life threatening in the high risk morbidly obese patient and may require prompt aggressive surgical excision.

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