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The diagnostic and surgical challenges of massive localized lymphedema



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Abstract

BACKGROUND: Massive localized lymphedema (MLL) is a rare entity first described in 1998 in patients with morbid obesity; the incidence is rising with the increased prevalence of morbid obesity. This report defines the clinical presentation and surgical challenges in 6 patients with MLL.

METHODS: The MLL in 6 patients with morbid obesity (weight range 270 to 585 lbs) involved the thigh in 3 patients, the calf in 1 patient, and the abdomen in 2 patients. The time from onset to presentation averaged 3 years (range 1 to 8 years). Two thigh lesions precluded ambulation because both legs could not be on the ground simultaneously; the 2 abdominal lesions were too heavy to permit ambulation.

RESULTS: The surgical excision required the use of pulleys to elevate the MLL tissues, which, on excision, weighed between 24 and 78 lbs. A long oval horizontal incision and a long transverse incision were used for the 2 abdominal lesions. Long horizontal oval limb incisions with multiple perpendicular cross incisions had to be used to excise MLL in the 4 limb lesions. In 2 cases, the vessel-sealing device was employed successfully for dissecting subcutaneous edematous tissue. Loose wound closure permitted postoperative lymph leakage, which continued for 3 to 8 weeks. The histology demonstrated fibrotic lymphatic tissue with vascular and lymphatic proliferation and edema; all patients did well.

CONCLUSIONS: MLL is rare and is best treated by surgical excision facilitated by pulleys and imaginative incisions to obtain primary closure. Long-term follow-up is necessary to assess for subsequent liposarcoma or angiosarcoma.

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Massive localized lymphedema (MLL) was first described as a distinct clinical entity by Farshid and Weiss¹ in 1998. MLL is an extremely large, pendulous, localized, and benign overgrowth of lymphoproliferative tissue typically seen in morbidly obese patients^{2,3} and can be easily confused grossly with gigantic liposarcomas.² Infrequently, it degenerates into a sarcoma if untreated for a long period of time. Shon et al⁴ reported a series of 5 patients where angiosarcoma arose in the background of MLL. Histologic

examination, usually, shows mature fat mixed with fibroblastic proliferation and vascular proliferation.² These lesions may grow to such gigantic proportions that ambulation becomes impossible because of the massive weight or, if located in the lower extremity, because of the protrusion of the lesion against the contralateral lower extremity so that both feet cannot be on the ground simultaneously. The lymphedema is well localized, a distinguishing feature from generalized-dependent lymphedema secondary to deep venous thrombosis, venous insufficiency, or venous hypertension. Surgical excision is the optimal treatment for MLL because, unlike classic lymphedema, it is localized and is irreversible; conservative treatment alone with antibiotics for associated cellulitis and compression dressings is doomed to failure.³ This report describes the clinical findings and surgical challenges in 6 MLL patients treated from 2003 to 2013.

Clinical Material

Between 2003 and 2013, 1 male and 5 female patients with MLL were treated. This project was approved by the Institutional Investigational Review Board because all the patients are deidentified. All patients were morbidly obese ranging from 330 (5'10") to 588 lbs (5'8"). The distribution of the lesions included 2 abdominal, 3 thigh, and 1 calf. The skin overlying the area of MLL was very thick, indurated, and uniformly exhibited peau d'orange that clinically mimics a liposarcoma. Moisture over the MLL was common. The skin surrounding the area of MLL was also edematous but did not show the overlying peau d'orange and brawny discoloration. The weight of the excised tissue ranged from 24 lbs in the patient with calf MLL to 78 lbs in one abdominal MLL patient. Prior surgical consultations typically led to recommendations of dietary weight loss and compression dressings.

All 6 patients presented with a long history of MLL, which had been gradually increasing in size from 3 years in the calf lesion to almost 10 years in the patients with abdominal wall lesions. The technical challenges of resecting these lesions are described in the following 3 brief case reports.

Patient #1

This 38-year-old super obese (body mass index > 60) male patient presented with MLL of the left thigh. It progressively got worse for the last 5 years. He presented in a wheelchair because the huge mass prevented him from getting both legs in a walking position. Surgical excision was recommended. The weight of the lesion was prohibitive for the surgical team, so a pulley system was utilized to elevate this huge mass during operation (Fig. 1). The size and shape of the lesion precluded excision by way of a simple oval incision; the 2 sides of the initial longitudinally oriented oval incision had to be tee'd off at 90° across the



Figure 1 This MLL of the left thigh weighed 58 pounds and was supported by a system of pulleys to facilitate resection.

thigh in a lateral direction at 5 different sites. The massive fatty tissue leading to the MLL was divided by using the vessel-sealing device starting circumferentially and then working down to the healthy adipose tissue, which covered the underlying muscles. Once the bulky lesion was excised, the wound was closed using interrupted polyglactin 910 sutures for the subcutaneous fascia and interrupted #2 nonabsorbable, polyester sutures placed far back from the primary wound and the tee'd off extensions to prevent wound dehiscence. These larger sutures were placed at least 3 cm beyond the wound margins. Between the large sutures, 3-0 nylon sutures, which likewise were placed far back from the skin edges, approximated the skin. On post-operative day 1, the patient was able to walk for the first time in many months and was discharged home on day 3 for home physiotherapy and frequent dressing changes to accommodate wound lymph leakage, for the next 6 weeks. The wound was completely healed by 3 months. The resected specimen weighed 58 lbs. The histology demonstrated excessive fibrous tissue associated with diffuse lymphedema and both lymphatic and vascular proliferation without evidence of malignancy.

Patient #2

This 45-year-old super obese (body mass index > 80) female patient presented with abdominal wall MLL. It had been growing for several years, now prohibiting walking because of the weight. She also presented to the surgical office in a wheelchair (Fig. 2). Excision was recommended. The operative approach was similar to that used for panniculectomy with a transverse oval incision. A pulley system had to be devised to elevate the huge segment of MLL. A 2-team approach was used with each team working from the side toward the middle. Despite this approach, the total operative time was almost 3 hours. As seen in all 6 patients, the fatty subcutaneous tissue adjacent to the MLL was

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