Massive Localized Abdominal Lymphedema Treatment Challenges

Khaled Al-Tarrah, MBchB, MRCS*†
Tarek Ashour, MBchB, EBOPRAS, MD*
Rajashankar Rao, MBBS, DA, FRCA‡
Sukhbir Rayatt, FDS, FRCS (Plast)*
Sunil S. Thomas, MS, FRCS (Plast)*

Summary: Massive localized lymphedema of the abdomen is a rare condition resulting from a neglected lower abdominal pannus associated with significant disability and morbidity. Compared to other surgical procedures, postbariatric surgery is usually considered a financial drain. In the United Kingdom, this requires National Health Service approval and delays may lead to sequelae that adversely impact on patients’ quality of life with increased morbidity. We present a wheelchair-bound patient whose body mass index increased from 53 to 82, while awaiting funding approval increasing her anesthetic and surgical risks. A multidisciplinary approach is mandatory for preoperative, intraoperative, and postoperative care for these patients including anesthetic input and high dependency unit care. Managing this patient was a significant anesthetic and surgical challenge with 47-kg resected tissue. The planning and perioperative measures to minimize morbidity are discussed. (Plast Reconstr Surg Glob Open 2020;8:e2601; doi: 10.1097/GOX.0000000000002601; Published online 17 January 2020.)

Massive localized lymphedema (MLL) is a rare condition observed in the morbidly obese populations. The epidemiology of MLL is currently unknown, but due to increasing prevalence of obesity worldwide, its incidence is expected to rise. MLL is associated with disturbance of efferent lymphatic flow possibly due to extensive adipose tissue. Other possible causes include previous surgery, trauma, and hypothyroidism. However, the pathogenesis of MLL remains to be fully elucidated. Therefore, diagnosing MLL can be clinically, pathologically, and radiologically challenging as these soft tissue lesions can mimic malignancy. Multiple published reports describe oncological resection of MLL as patients usually present with large masses that radiologically and histologically mimic liposarcoma or lymphosarcoma, hence MLL has been termed “pseudosarcoma.” Also of concern are reports identifying MLL as a risk factor for the development of angiosarcoma.

Depending on clinical history, anatomical site, examination, and investigations, MLL can be managed conservatively or surgically. Conservative measures include antibiotics, for associated cellulitis and complete decongestive therapy. Surgery is advocated when there is functional deficit, including immobility, recurrent infections, and suspicion of malignancy.

MLL has been reported to affect various anatomical sites including thigh, popliteal fossa, scrotum, mons pubis, and abdomen. Relevant to this report, abdominal MLL is associated with significant functional disabilities of mobilization, back pain, skin intertrigo, suprapubic edema, and poor local hygiene. Abdominal dermolipectomy is the recommended surgical procedure, although it is considered a cosmetic procedure by most institutions. In the United States, this operation may be covered by medical insurance, but in the United Kingdom it is done in the private sector or with funding approval on the National Health Service (NHS).

With Individual Funding Request approval, our patient was treated on the NHS. The process took about a year to complete, further increasing her morbidity. The perioperative planning is discussed with a satisfactory outcome and further plan for gastric banding. To the best of our knowledge, this is the largest abdominal debulking procedure reported in the United Kingdom.
CASE REPORT

A 51-year-old white woman with the history of hypertension, vitamin D deficiency, and severe osteoarthritis presented with morbid obesity and concerns about an overhanging pannus which had progressively increased over 8 years with significant deterioration of her personal care, hygiene, and overall quality of life (QOL). She was wheelchair bound and barely managed to mobilize from her bed with crutches.

She was referred to Plastics after a failed laparoscopic gastric sleeve procedure due the excessive thickness (approximately 6 cm) of the anterior abdominal wall. She was morbidly obese with a large pannus extending down to her lower legs with peau d’orange appearance and ulceration [body mass index (BMI): 53; height: 1.57 m; weight: 133 kg]. A lower abdominal apronectomy was planned, without umbilical preservation on the NHS. The funding approval took over a year during which time the patient gained 69 kg (weight: 202 kg; BMI: 82) with deterioration in mobility, personal care, and “morale” (Fig. 1).

The apron now reached her ankles with worsening lymphedema and mobilization. In addition to surgery, she had significant anesthetic risks such as deep vein thrombosis, myocardial infarction, and pulmonary embolism. A computed tomography (CT) scan confirmed a 14 cm × 7 cm midline rectus defect with no bowel herniation. She required a bariatric bed, specialized operating table with extensions, and postoperative high dependence unit care. Preoperative planning included deep vein thrombosis prophylaxis with daily 9,000-U tinzaparin, thromboembolic deterrent stockings, and Flowtron pumps.

The surgery involved 2 teams to minimize anesthesia duration and surgical risks. The apron en bloc excision was started superiorly above the umbilicus till the suprapubic area was reached over the rectus sheath. This was done to reduce the time, and the apron was then lifted supero-laterally, so minimizing the burden on the patient’s vitals and respiration. The major blood vessels identified in the CT scan, including large superficial inferior epigastric veins (2.7 cm diameter), were preemptively ligated and rectus sheath defect plicated using a double running 2/0 polydioxanone suture. The umbilicus sacrificed to complete the lower incision. The excision was completed removing a 47-kg abdominal pannus. The wound was closed in layers with quilting, fibrin sealant, and vacuum assisted closure drains without undermining to minimize wound-healing complications (see figure, Supplemental Digital Content 1, which displays preoperative transverse CT scans of patient demonstrating the size of superficial inferior epigastric vessels, http://links.lww.com/PRSGO/B278) (see figure, Supplemental Digital Content 2, which displays intraoperative clinical photography of excised surgical pannus, http://links.lww.com/PRSGO/B279) (see figure, Supplemental Digital Content 3, which displays intraoperative clinical photography of surgical defect following excision of pannus, http://links.lww.com/PRSGO/B280).

The abdominal wound was closed in multiple layers with quilting polydioxanone sutures and fibrin glue (ARTISS, Baxter, UK), to prevent collection. Two vacuum assisted closure drains on each side were used on 80 mm Hg intermittent pressure to further prevent fluid collection (RENASYS; Smith & Nephew, London, UK). She mobilized early with physiotherapy and was discharged on the 17th postoperative day. She had 2 areas of minor wound breakdown which healed with dressings. Three months postoperatively, she weighed 149 kg (BMI: 60) and was able to mobilize with minimal support (Fig. 2).

DISCUSSION

Morbid obesity is a complex multifactorial problem that adversely affects an individual’s health and QOL. It also impacts morbidity and increases the risk for malignancy, cardiovascular disease, diabetes, osteoarthritis, and higher all-cause mortality. This is concerning, as obesity prevalence is increasing worldwide, with an economic burden and increasing annual total health-care costs by...
36%,

In the United Kingdom, most bariatric procedures are perceived as “cosmetic” and therefore not routinely funded on the NHS, without a specific Individual Funding Request approval. This took more than a year for our patient resulting in gaining 68 kg, significantly increasing anesthetic and surgical risks, including difficult intubation, airway management, cardiorespiratory compromise, reduced lung compliance, increased ventilation-perfusion (V/Q) scan mismatch, and systemic and pulmonary hypertension.

Intravenous access is difficult for drug administration, and adipose tissue influences the pharmacokinetics of many drugs and general anesthetic agents.

Intraoperatively, there is an increased risk of deep vein thrombosis, myocardial infarction, pulmonary embolism, and stroke. Meticulous preoperative planning and efficient surgical execution are imperative to minimize complications and morbidity. A 2-team approach also reduces the operating/anesthetic time.

Standard apronectomy involves an incision in the patient’s natural suprapubic crease that extended laterally toward and beyond the anterior superior iliac spine. The incision is then deepened down to the musculoaponeurotic plane, which is followed superiorly. The apron is then excised and umbillectomy performed if needed. Sachs and Murray have published an overview of panniculectomies in detail. This classical approach may not be feasible in extreme cases such as abdominal MLL, where apronecetomy/panniculectomy carries technical challenges. Various measures have been described to lift the “apron,” including overhead cranes, Steinmann pins with pulleys, and hydraulic lifts.

In our patient, these were considered unsafe by medical engineering. An extension to the operating table was used to abduct the legs to rest the apron.

Weathers et al reported a 94-kg apron resection. In the United Kingdom, weights of 25.4 and 35 kg were reported. This patient with 47-kg resection would possibly be the largest apron resection reported till date in the United Kingdom.

Obesity is associated with multiple postoperative complications including wound infection and dehiscence. Koulaouzidis et al reported 37% incidence of wound breakdown following apronecetomy. Manahan and Shermak also reported 20% wound-related complications following massive panniculectomy. Our patient had a minor wound breakdown which healed with dressings.

CONCLUSIONS

MLL is a challenging surgical problem due to a neglected abdominal apron that merits early recognition and intervention. Surgical management needs careful multidisciplinary perioperative planning. Delay increases risks further worsening mobility and morale. Adjuncts of quilting sutures, use of fibrin glue, and vacuum assisted closure dressings also reduce complications. The improvement in the patients’ QOL more than compensates for the initial surgical cost including benefits by reduction in ongoing nursing and social care expenses.

Khaled Al-Tarrah, MBchB, MRCS
Institute of Inflammation and Ageing
Birmingham University Medical School
Birmingham B15 2TT, UK
E-mail: kxa455@bham.ac.uk

ACKNOWLEDGMENT

The authors would like to thank Department of Clinical Photography and Medical Illustration for their assistance and help.

REFERENCES

1. NCD Risk Factor Collaboration (NCD-RisC). Trends in adult body-mass index in 200 countries from 1975 to 2014: a pooled analysis of 1698 population-based measurement studies with 19.2 million participants. Lancet. 2016;387:1377–1396.

2. NCD Risk Factor Collaboration (NCD-RisC). Worldwide trends in body-mass index, underweight, overweight, and obesity from 1975 to 2016: a pooled analysis of 2416 population-based
measurement studies in 128.9 million children, adolescents, and adults. *Lancet*, 2017;390:2627–2642.

3. Manduch M, Oliveira AM, Nascimento AG, et al. Massive localised lymphoedema: a clinicopathological study of 22 cases and review of the literature. *J Clin Pathol* 2009;62:808–811.

4. Wu D, Gibbs J, Corral D, et al. Massive localized lymphedema: additional locations and association with hypothyroidism. *Hum Pathol* 2000;31:1162–1168.

5. Farshid G, Weiss SW. Massive localized lymphedema in the morbidly obese: a histologically distinct reactive lesion simulating liposarcoma. *Am J Surg Pathol* 1998;22:1277–1283.

6. Porro J, Walsh J. Massive localized lymphedema of the thigh mimicking liposarcoma. *Radiol Case Rep* 2016;11:391–397.

7. Kotidis E, Cepaiytye D, Petrikas G, et al. Massive localized lymphedema in the morbidly obese patient: a clinical entity mimicking liposarcoma. *Wounds* 2015;27:249–252.

8. Goshtasby P, Dawson J, Agarwal N. Pseudosarcoma: massive localized lymphedema of the morbidly obese. *Obes Surg* 2006;16:88–93.

9. Salas S, Stock N, Stoeckle E, et al. Chronic lymphedema due to morbid obesity: an exceptional cause of abdominal wall angiosarcoma. *Virchows Arch* 2008;453:217–219.

10. Azam M, Saloojaan H, Bierlig S, et al. Cutaneous angiosarcoma complicating morbid obesity. *Arch Pathol Lab Med* 2001;125:531–533.

11. Shehan JM, Ahmed I. Angiosarcoma arising in a lymphedematous abdominal pannus with histologic features reminiscent of Kaposi’s sarcoma: report of a case and review of the literature. *Int J Dermatol* 2006;45:499–503.

12. Shon W, Ida CM, Boland-Fromming JM, et al. Cutaneous angiosarcoma arising in massive localized lymphedema of the morbidly obese: a report of five cases and review of the literature. *J Cutan Pathol* 2011;38:560–564.

13. Chopra K, Tadisina KK, Brewer M, et al. Massive localized lymphedema revisited: a quickly rising complication of the obesity epidemic. *Ann Plast Surg* 2015;74:126–132.

14. Fife C. Massive localized lymphedema, a disease unique to the morbidly obese: a case study. *Ostomy Wound Manage* 2014;60:30–35.

15. Jabbar F, Hammoudeh ZS, Bachusz R, et al. The diagnostic and surgical challenges of massive localized lymphedema. *Am J Surg* 2015;209:584–587.

16. Saty S, Pandya S. Should a panniculectomy/abdominoplasty after massive weight loss be covered by insurance? *Ann Plast Surg* 2008;60:502–504.

17. Gurunluoglu R. Panniculectomy and redundant skin surgery in massive weight loss patients: current guidelines and recommendations for medical necessity determination. *Ann Plast Surg* 2008;61:654–657.

18. https://www.england.nhs.uk/publication/specialised-services-individual-funding-requests//. Accessed March 13, 2019

19. Vegel AJ, Shah N, Lidor AO, et al. Patient-reported quality of life after bariatric surgery: a single institution analysis. *J Surg Res* 2017;218:117–123.

20. Vischer TL, Seidell JC. The public health impact of obesity. *Annu Rev Public Health* 2001;22:555–375.

21. Taylor VH, Forhan M, Vigod SN, et al. The impact of obesity on quality of life. *Best Pract Res Clin Endocrinol Metab* 2013;27:139–146.

22. Seidell JC, Halberstadt J. The global burden of obesity and the challenges of prevention. *Ann Nutr Metab* 2015;66 Suppl 2:7–12.

23. Flegal KM, Kit BK, Orpana H, et al. Association of all-cause mortality with overweight and obesity using standard body mass index categories: a systematic review and meta-analysis. *JAMA* 2013;309:71–82.

24. Kent S, Fusc F, Gray A, et al. Body mass index and healthcare costs: a systematic literature review of individual participant data studies. *Obes Rev* 2017;18:860–879.

25. Dobbs R MJ, Woetzl J, Savers C, et al. How the world could better fight obesity. Available at: https://www.mckinsey.com/industries/healthcare-systems-and-services/our-insights/how-the-world-could-better-fight-obesity. Accessed July 27, 2018

26. Nelson G, Clayton R. Anaesthesia in the obese patient. *Anaesthesia & Intensive Care Medicine* 2017;18:472–476.

27. Lotia S, Bellamy MC. Anaesthesia and morbid obesity. *Continuing Education in Anaesthesia Critical Care Pain* 2008;8:151–156.

28. Sachs D, Murray J. Panniculectomy. Available at: https://www.ncbi.nlm.nih.gov/books/NBK499822/. Accessed August 3, 2019

29. Töth T, Chang ChienYC, Kollár S, et al. Massive localized lymphedema arising from abdominal wall: a case report and review of the literature. *Case Rep Pathol* 2015;2015:375909.

30. El-Sharkawy MS, Al-Rikabi AC, AlArfaj N, et al. Localised massive lymphedema masquerading as an anterior abdominal mass mimicking a liposarcoma. *Am J Med Sci* 2014;347:84–85.

31. Evans C, Deborj J, Howe H, et al. Massive panniculectomy results in improved functional outcome. *Am J Surg* 2014;207:444–4; discussion 444.

32. Felmerer G, Karcz W, Földi E, et al. Integrated concept of treatment for reduction of morbidity after resection of panniculus morbidus associated with lymphoedema. *J Plast Surg Hand Surg* 2012;46:172–176.

33. Koulaxouzidis G, Goerke SM, Eisenhardt SU, et al. An integrated therapy concept for reduction of postoperative complications after resection of a panniculus morbidus. *Obes Surg* 2012:22:549–554.

34. Ollapalli J, Koong D, Panchacharavel G, et al. New method of abdominoplasty for morbidly obese patients. *ANZ J Surg* 2004;74:504–506.

35. Warner RM, Smyrou GE, Foureier L. A case of apronecrotomy for treatment of massive abdominal lymphoedema. *European Journal of Plastic Surgery* 2003;26:260–262.

36. Moon Y, Pyon JR. A rare case of massive localized lymphedema in a morbidly obese patient. *Ann Plast Surg* 2016;74:125–127.

37. Weathers WM, Wolfswinkel EM, Rebouse R, et al. Suspension of panniculus morbidus: a salvage procedure with a steep learning curve. *Plast Reconstr Surg* 2008;121:108–114.

38. Petty P, Manson PN, Black R, et al. Panniculus morbidus. *Ann Plast Surg* 1992;28:442–452.

39. Manahan MA, Shermak MA. Massive panniculectomy after massive weight loss. *Plast Reconstr Surg* 2011;128:786–787.

40. Friedlich JB, Petros RV, Askay SA, et al. Resection of panniculus morbidus: a salvage procedure with a steep learning curve. *Plast Reconstr Surg* 2012;129:1171–1179.

41. Fadel MG, Chatzikonstantinou M, Gilchrist C, et al. Panniculus morbidus: obesity-related abdominal wall lymphoedema. *BMJ case reports* 2017;2017:bcr-2016-219023. doi:

42. Goldberg C, Fish J. Helpful surgical technique in massive abdominal panniculectomy. *Canadian Journal of Plastic Surgery* 2001;9:205–207.

43. Doyle SL, Lysaght J, Reynolds JV. Obesity and post-operative complications in patients undergoing non-bariatric surgery. *Obes Rev* 2010;11:875–886.

44. Cohen RV, Luque A, Junqueira S, et al. What is the impact on the healthcare system if access to bariatric surgery is delayed? *Obes Relat Dis* 2017;13:1619–1627.