Case Report: Lymphedema Lower Limb-Surgical Management

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Abstract: Lymphedema may be defined as abnormal limb swelling caused by accumulation of increased amounts of high protein ISF secondary to defective lymphatic drainage in presence of (near) normal net capillary filtration(1). At birth, 1 in 6000 people will develop lymphedema with overall prevalence of 0.13-2%(1). Despite significant impact on quality of life, many do not seek medical advice because of embarrassment and belief that nothing can be done(1). Here we report a case of successful reduction in volume of lymphedema limb by Charles operation.

Keywords: Lymphedema; Charles operation; Surgical management

I. Introduction
Lymphedema may be defined as abnormal limb swelling caused by accumulation of increased amounts of high protein ISF secondary to defective lymphatic drainage in presence of (near) normal net capillary filtration(1). The condition is not only associated with significant physical symptoms and complications but is also a frequent cause of emotional and psychological distress, which can lead to difficulties with relationships and work(1). Hence it not only disfigures, also it is disabling and distressing. At birth, 1 in 6000 people will develop lymphedema with overall prevalence of 0.13-2%(1). Lymphedema may be primary or secondary. Lymphedema refractory to non operative management may require surgical treatment(2). Principle of excisional surgery is to remove excess tissue to decrease the volume of extremity(3). Lymphedema is often misdiagnosed and mistreated by doctors, who frequently have a poor understanding of the condition, believing it to be primarily a cosmetic problem(1). Early diagnosis is important because relatively simple measures can be highly effective and disabling late disease often very difficult to treat(1). It occurs more frequently in females and peaks between 12-16 years of age(4). Most common cause of secondary acquired lymphedema universally, is parasitic infection by filariasis(5). The disease has a vast clinical spectrum from limited partially pitting slowly progressive swelling of distal end of extremity to severe and complicating chronic edema with fibrotic change(6) and consequently the non pitting type of whole limb causing limb heaviness, restricted movements and resultant disability same as our case(7). Surgical correction is required when conservative treatment fails, and methods available are bypass procedures, liposuction and reduction procedures(1).

II. Case Report
The patient was a 44 years old female hailing from Alappuzha who is unmarried came with non healing ulcer for 6 years and suffering from bilateral lower limb edema for the past 25 years. History of recurrent ulcer in the limb with cellulitis and oozing and multiple hospitalisation for the same. For the past two years she was almost bedridden due to heaviness of the limb. Comparing to the left limb, right limb is more bulky. She had no comorbidities. History of filarial lymphedema for her father and carcinoma breast for her sister. On examination significant increase in girth of both lower limb starting from the thigh segment more on the right side. Skin is thickened and hyperkeratotic with multiple non healing ulcers and multiple papillomatous lesions. Malignancy had been excluded by skin and subcutaneous tissue biopsy and the peripheral vascular disease excluded by doppler examination. Preoperatively patient evaluated with MRI examination to rule out chronic venous insufficiency. Ulcer had been treated with antibiotics and daily cleaning and dressing. Patient was prepared for surgery and debulking of medial segment of upper thigh region was planned. Patient placed in supine position over 2 operative tables since 1 table could not accommodate her. Excision of the thickened skin and fibrotic subcutaneous tissue was done up to deep fascia level. The resultant defect closed in layers. Nonpostoperative complications like haematoma formation, wound infection or wound break down occurred. The thigh circumference reduced from 120 cms to 90 cms. Patient stood on 3rd postoperative day and started walking on 5th postoperative day where our team felt like our own kid started walking from crawling. On 12th postoperative day sutures were removed and the patient discharged to home.

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III. Discussion

Lymphedema is defined as deficiency in lymphatic system with protein rich fluid accumulated in interstitial space. It may be primary or secondary. Secondary lymphedema commonly due to filarial parasitic infestation(8). Recurrent cellulitis could be both the cause and complication of secondary lymphedema. Repeated infection and inflammation may destroy the lymphatic vessels, leading to lymphedema, the protein rich lymph serve as a medium providing microbial proliferation. Besides repeated skin infection and affecting quality of life, another rare but severe complication of chronic lymphedema was the malignant transforming to lymphangiosarcoma classically seen in postmastectomy lymphoedematous arm(Stewarts Treves Syndrome) but has also reported in primary lymphedema and chronic filarial lymphedema. The tumour has very poor prognosis and the only treatment is amputation. Symptoms and signs are swelling, buffalo hump, stemmer’s sign, fibrosis, hyperkeratosis and ulceration(1). Clinical classification called Burner’s grading ranging from subclinical,I,II, and III. Diagnosis is by CT and MRI imaging(8). For patients with edema of unknown cause and a suspicion of lymphedema, lymphoscintigraphy is the diagnostic test of choice(8). Even though lymphoscintigraphy cannot differentiate between primary and secondary lymphedema, it has sensitivity of 70-90% and specificity of nearby 100% in differentiating lymphedema from other causes of limb swelling. Direct contrast lymphangiography provides the finest details of lymphatic anatomy(9) but is used infrequently and reserved for preoperative evaluation of selected patients who are candidate for direct operations on their lymphatic vessels(8). The most promising new test appears to be contrast magnetic resonance lymphangiography, which is performed after intracutaneous injection of gadobenate dimeglumine into interdigital webs of dorsal foot. Reported data suggest that the new test is capable of visualising the anatomy and functional status of lymph flow transport of lymphatic vessels and lymph nodes of lymphoedematous limb(8). Majority of lymphedema patient can be treated with combination of limb elevation, a high quality compression garment, complex decongestive physical therapy, and compression pump therapy, and operative treatment may be considered for the patients with advanced complicated lymphedema for whom management with non operative means has failed(8). There are mainly two types of surgery: Bypass surgery and Resection procedures. Bypass surgery creates anastomosis between lymphatic ducts or nodes and vein and is more indicated for early stage lymphedema and it attempts to correct underlying cause directly. Charles procedure was first described in 1912 with radical resection of all affected skin and subcutaneous tissue down to deep fascia with coverage using split thickness skin grafts harvested from excised specimen. Because it focuses on removal of excess tissue instead correcting underlying pathology, the edema may return. Charles procedure is a significant debulking surgery and some complications may develop after this procedure, including massive blood loss that needs transfusion, poor graft patency that needs regrafting, continuous lymphatic fluid oozing, post operative infection, unsatisfactory cosmetic results, hypertrophic scar, papillomatosis, recurrent cellulitis, and lymphatic fistula. Several modified methods has been described such as delayed grafting, the donor site choosing, combination use with negative pressure dressing and excision with preservation of perforators.

Figure 1: MRI showing cut section of (R) thigh. Figure 2: Pre-excision and Post-excision limb.
IV. Conclusion

Our case demonstrates that resection technique can be modified to a more cosmetic and volume reducing one with less disfiguring scars, better functional results, low rate of wound complications. One step from our side (in form of strong thought to do something to the patient like my chief had) can make these patients walk miles through (literally).

References