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Case Report

Charles procedure combined with staged subcutaneous excision in treating a case of chronic massive lymphedema: a case report

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ABSTRACT

Lymphedema in its advanced stage is a physically and psychologically debilitating condition as conservative management alone in this condition, barely makes any difference in patient's quality of life. Here we present a case report where we were able to successfully treat our patient of massive lymphedema and help him walk on his feet, by an approach where we combined Charles procedure and staged subcutaneous excision of soft tissue.

Keywords: Lymphedema, Subcutaneous excision, Charles procedure, De-bulking surgery

INTRODUCTION

As defined by the WHO, peripheral lymphedema is the clinical manifestation of insufficient and deranged lymphatic transport. The cause for this can be primary or secondary.[1]

Congenital lymphatic dysplasia is the most common condition leading to primary lymphedema but is relatively less common as compared to secondary. Filariasis, a disease caused by the parasite *Wuchereria bancrofti*, is the most common cause of secondary lymphedema worldwide, while in the developed countries, lymph node resection, radiation therapy, metastasis, direct trauma is some of the most common causes.[2]

The fundamental pathology in lymphedema is an inadequate lymphatic drainage either due to mechanical insufficiency or dynamic insufficiency the ultimate consequence of which is the progressive accumulation of protein rich fluid in the interstitial space.[1] This protein rich lymph triggers an inflammatory response which induces irreversible proliferation of parenchymal and stromal elements with lipogenesis, and connective tissue overgrowth, ultimately giving rise to a swollen and deformed limb with decreased functionality.[3]

Lymphedema can be staged based on the physical condition of the extremities as follows:[1]

Stage 0: latent or subclinical condition where limb changes are not evident despite impaired lymph transport.

Stage 1: Early accumulation of protein rich fluid which subsides with limb elevation. Pitting present

Stage 2: More limb changes with proliferation of subcutaneous fat and fibrosis, limb elevation alone doesn't reduce tissue swelling, and pitting may or may not be.

Stage 3: Includes lymph static elephantiasis with absent pitting, trophic skin changes such as acanthosis, increased skin thickness, massive deposition of fat and fibrosis, and warty overgrowths.

The management of lymphedema can be by non-operative or operative methods[1]

Non operative treatment is feasible in early stages of lymphedema and the options include:

Physical therapies like complete decongestive therapy (CDT) which involves manual lymphatic drainage, skin care and muscle pumping exercises, use of compression stockings, limb physiotherapy, drug therapy-anti microbial and anti-filarial treatment, psychosocial rehabilitation to improve the quality of life of lymphedema patients.

Surgical management is required in late stage (III) and is always used in adjunct with life-long non operative methods.

Microsurgical procedures: These are the techniques which attempt to re-establish the normal lymphatic physiology and include derivative method like lympho-venous anastomoses (LVA) or reconstructive methods like lymphatic collector or an interposition vein segment to restore lymphatic continuity.

Vascularized lymph node transplantation where in healthy superficial lymph nodes from an uninvolved area is transplanted to the diseased site together with the vascular supply.

Liposuction is another surgical method which is useful in cases of lymphedema with excess fat deposition and less fibrosis but have failed to respond to non-operative management.

Surgical resection is the mainstay of treatment in most severe forms of fibrosclerotic lymphedema. It is a palliative procedure which focuses on decreasing the limb volume to improve its functional status. The Charles procedure, first described in 1912, involves skin and soft tissue excision till the level of the deep fascia followed by split thickness skin grafting, from the excised specimen to augment wound closure. On the other hand, de-bulking technique involves subcutaneous excision of soft tissue and skin is preserved, hence does not require skin grafting.[3-6]

CASE REPORT

A 44-year-old male patient, hailing from Uttar Pradesh, but a resident of Surat, Gujarat, street vendor by occupation, was brought by his son to our surgical OPD, with a massively enlarged right lower limb, gradually progressive for 20 years. As per the history given by patient, he had previously sought medical consultations at different places several times, but was only conservatively managed with compression stockings, and oral medication. But now, as the condition had progressed to the extent of becoming dependable for his day-to-day activities, he wanted a permanent solution for his condition. He had no significant past history, and had no existing comorbidities. On physical examination, patient had unilateral right sided lower limb enlargement, extending from foot till the mid- thigh, massive to the extent of masking his toes, foot, ankle joint and knee joint. Based on history and physical examination, diagnosis of stage III lymphedema was made, and because of extensive fibrosis and skin changes, surgical resection was planned for the patient after getting pre-operative anaesthetic clearance.

Stage 1 surgery*:* The Charles procedure and de-bulking were combined in the first surgery performed on our patient. Fibrosed overgrowth on the foot was completely excised leaving a raw area behind which was covered by skin grafting, for which skin was harvested from the excised specimen. In the same sitting, connective tissue growth in the ankle region was removed by subcutaneous excision with primary skin closure. Negative suction drain was placed to avoid any immediate subcutaneous collection.

Suture site and STG site dressings were opened on the 3rd post- operative day, to see a healthy wound and a graft that was accepted.

Emergency surgery*:* Progressive blackening of skin was seen around the suture site by 5th post- operative day with sero-haemorrhagic discharge which led to give away of sutures and wound gaping. Patient had to be taken to emergency surgery for debridement of the suture site on post-operative day 5, following which wound was kept open without suturing.

Daily dressing of the wound was done for next 7 days and and after ensuring a healthy wound, patient was posted for second stage debulking.

Stage 2 surgery*:* Second stage de-bulking was done after 12 days of the first surgery. Large growths on the medial and lateral aspects of the lower limb, below knee were excised subcutaneously, and a primary skin closure was done. The lateral skin incision was an extension of previous ankle site incision, and hence while closure, the ankle site (which had been debrided and kept open previously) skin closure was also done in the same setting after placing negative suction drain. Medial part of ankle site wound was wide enough to require STG, and was retrieved from the excised specimen.

Dressing was opened on 3rd post-operative day, and drain removed on 5th post-operative day. Patient was trained in supported mobilization using walker, 5th post-operative day onwards. During the entire period post operatively, decongestion of the operated limb was ensured by tight crepe bandage application, limb elevation, physiotherapy exercises together with once daily dressing of suture site done under strict aseptic precautions and was discharged on the 10th post-operative day after suture removal. Patient who was brought on stretcher by his son, on the day of admission, went home walking with the help of walker on discharge.

Figure 1: Pre-operative anterior view of right lower limb lymphedema before stage 1 surgery.

Figure 2: Preoperative posterior view of right lower limb lymphedema before stage 1 surgery.

Figure 3: Intraoperative image of stage 1 surgery, Charles procedure and de-bulking were combined here.

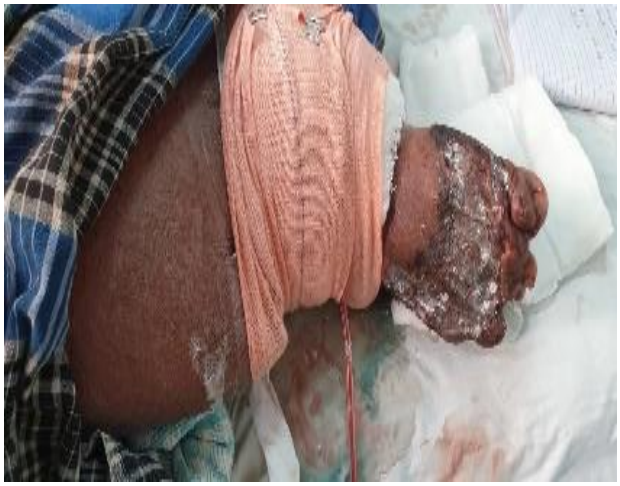

Figure 4: Post-operative day 3 image after stage 1 surgery, graft accepted.

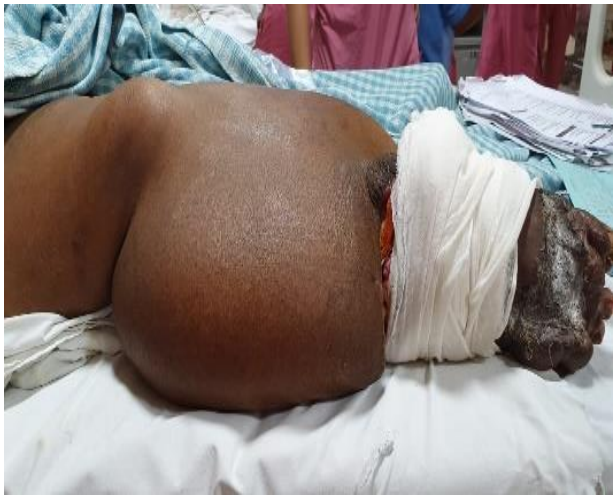

Figure 5: Immediate post-operative image after emergency debridement of blackened suture site.

Figure 6: Immediate post-operative lateral view after stage 2 surgery. (A) Lateral extension of ankle site incision was closed by skin suturing. (B) Medial part of ankle site excision was closed by STG.

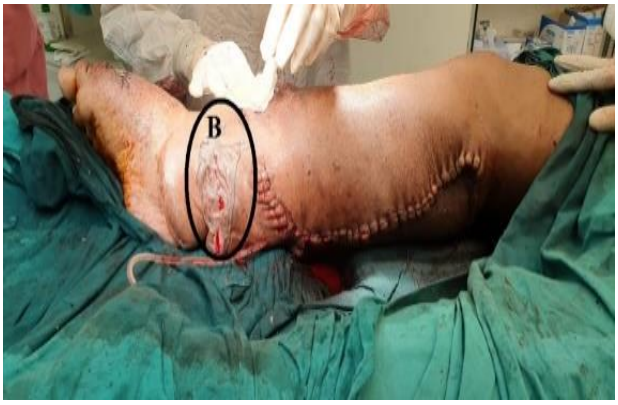

Figure 7: Immediate post-operative medial view after stage 2 surgery. (B) STG done on the medial aspect of ankle site incision.

Figure 8: Final outcome of right lower limb, on post-operative day 8 after stage 2 surgery.

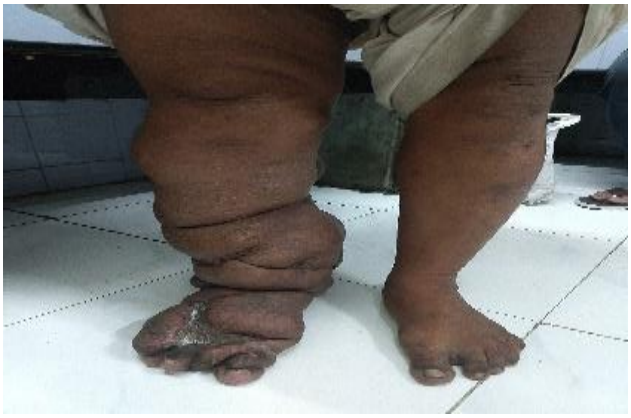

Figure 9: Picture taken at 6 months follow-up.

DISCUSSION

Our patient, being a case of advanced stage 3 lymphedema with excessive fibrosis, skin changes and functional incapacity, required surgical resection to reduce the size of massively enlarged limb so as to render him walkable, sufficient enough to lead an independent life. Considering the massive volume of the limb, surgery was planned in stages, so as to avoid complications like severe blood loss, and wound infection. The result of this was gratifying both to the patient and us, the surgeons.

The major goal of our treatment plan was to reduce the limb size together with preventing recurrent cellulitis of the involved limb due to skin infections. Hence throughout the hospital stay, patient was advocated proper limb hygiene, limb elevation, compression therapy using tight crepe bandaging, physiotherapy exercises, all of which basically aimed at decongestion of the involved limb. Special importance was given to educating the patient regarding skin hygiene techniques, by regular cleaning of skinfolds, creases and inter-digital spaces with soap water and rendering them dry using dry clothes, avoiding damp skin by regular mopping as moist skin attracts fungal infection. This had an added advantage of eliminating post-operative wound infection and graft rejection in our patient with minimal use of antibiotics post-operatively.

Surgical resection either by Charles method or by subcutaneous de-bulking is a traditional major operative technique which is avoided by most of the surgeons because of the high rate of morbidity and mortality associated with it, also because of the high chances of recurrence of lymphedema as the cause still persists even after surgery.[7] Likely, patient was denied surgery by many doctors at his previous consultations (as told by the patient himself). The strong decision made by our unit chief, to operate on this patient, together with the combined efforts of the entire unit under his valuable guidance, has today made the patient walkable and self-dependent.

CONCLUSION

Our case serves as an example for many surgeons, in boldly stepping ahead to opting for a relatively lesser chosen option in treating lymphedema, which can change a young patient's life for the better. Further, combining various surgical techniques of treating lymphedema together with aggressive non operative management can ultimately give unexpected gratifying outcomes with better functional results.

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