CASE REPORT

Double autologous lymph node transplantation (ALNT) at the level of the knee and inguinal region for advanced lymphoedema of the lower limb (elephantiasis)

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Received 16 August 2013; accepted 2 September 2013

KEYWORDS
Lymphoedema; Lymph node transplantation; Microsurgery; Lower limb

Summary Primary lower-limb lymphoedema is a chronic, progressive and debilitating condition with a difficult management, especially in advanced cases (elephantiasis). Recently, autologous lymph node transplantation (ALNT) appears to be a promising treatment for extremity lymphoedema. A case of a double ALNT for an advanced primary lower-limb lymphoedema is here reported: a contralateral inguinal lymph node flap was transferred to the knee and, in a second surgery, a thoracic lymph node flap was transplanted to the inguinal region. Clinical outcomes at 5 months postoperatively are very satisfactory with reduction in limb circumpresentences and improvement in skin quality and social impairment.

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Primary lower-extremity lymphoedema is difficult to manage and remains a challenging problem. The ideal treatment for lymphoedema of the limbs must restore function by controlling infection and reducing the swelling of the extremity, improve cosmetic appearance and improve the quality of life.

Physiotherapy (manual drainages, bandages and pressotherapy) has been classically the usual treatment for lymphoedema, but it aims to control the progression of the disease, with no curative purpose.
Microsurgical techniques for lymphoedema treatment such as autologous lymph node transplantation (ALNT) are gaining popularity as an alternative method to physiotherapy.1–9

Case report

A 57-year-old male patient, with a medical history of diabetes mellitus and hypertension, presented to our clinic with a primary right lower-limb lymphoedema. It was a tarda case, with an onset age of 52 years and a rapid progression to advanced stage (elephantiasis). Physical examination revealed a severe lymphoedema of the right lower extremity (Figure 1(A, B)), affecting the entire limb, but with more severity below the knee (elephantiasis of the leg). Skin changes such as discolouration, hyperkeratosis and papillomatosis were present. The patient also suffered from several repetitive episodes of lymphorrhoea, cellulitis and ulceration.

Lymphoscintigraphy showed a hypoplastic pattern with absence of lymph nodes and drainage.

Figure 1  (A, B) Primary lower limb lymphoedema, preoperative images; (C, D) Postoperative images at a follow up of five months.
Previous conservative treatment with physiotherapy (manual drainages, pressotherapy and bandages) was unsuccessful.

A surgical approach was decided on. An ALNT was performed using a contralateral inguinal lymph node flap based on branches from the superficial circumflex iliac vessels and containing superficial femoral nodes. The flap was transferred to the popliteal fossa and anastomosed to genicular artery branches and saphenous venous branches. The postoperative period was uneventful, with immediate improvement in perimetry of the limb and skin quality. Nevertheless, recovery was not complete, so that a second ALNT was necessary. A thoracic flap based on branches of the lateral thoracic vessels was placed at the level of the inguinal region and anastomosed to femoral artery branches and saphenous venous branches.

After the second surgery, there was a further reduction on circumferences of the limb, ulcers on the leg healed and skin quality improved (Figure 1(C, D)).

Postoperatively, the patient followed a physiotherapist management consisting of manual drainage, compressive bandages and compression garments. Perimetry of the limb was used as an indirect assessment of the volume. Standardised circumferential measurements were made preoperatively and 5 months postoperatively at five locations on the affected and normal limbs: dorsum of the foot, ankle, 12 cm below the knee (maximum circumference of the leg), knee and 10 cm above the knee (thigh). The reduction rate of the circumference of the lymphoedematous limb was defined as the preoperative difference between the circumferences of the affected and healthy limbs minus the postoperative difference and divided by the preoperative difference (Table 1).

Moreover, there was not any new episode of infection or ulceration on the affected limb, and the patient referred a better skin elasticity and texture with improvement in heaviness sensation and social impairment.

**Discussion**

This case report shows good results with the use of ALNT in advanced lymphoedema (elephantiasis) of the lower limb. The exact mechanism of action of the ALNT remains still unclear, but two processes are probably involved: an ‘internal pump’ and suction mechanism and lymphangiogenesis promotion.

Lymphatic nodes represent physiologic lymphaticovenous anastomosis. The lymph node transfer acts by means of an internal pump and suction mechanism: the pump is driven by the strong hydrostatic force into the flap from the donor artery, and the suction is continued by the donor vein via natural lymphaticovenous connections inside the flap. As the subcutaneous interstitial pressure decreases, more lymph is recruited from the surrounding tissue in a ‘catchment effect’. Human lymph nodes express high levels of vascular endothelial growth factor c (VEGF-c). Experimental lymphoedema models show that lymphatic vasculature has a high capacity for regeneration. Therefore, transfer of lymph nodes with endogenous lymphatic growth factor expression may promote lymphangiogenesis.

Most previous reports of ALNT discuss iatrogenic upper-limb lymphoedema, using typically superficial groin lymph node transplantation. Experimental lymphoedema models show that lymphatic vasculature has a high capacity for regeneration. Therefore, transfer of lymph nodes with endogenous lymphatic growth factor expression may promote lymphangiogenesis.

Alternative donor sites can be used for lower-limb lymphoedema, such as thoracic lymph nodes, submental nodes or supraclavicular nodes. Encouraging outcomes have also been reported with reduction in leg circumference with these flaps.

In severe lymphoedema, a complete cure may not be possible with ALNT alone. In these cases, as in the patient of the present report, a second flap can be performed, improving results. Additional procedures, such as ablative surgeries or liposuction, may also be required for further improvement in limb perimetry and cosmetic appearance.

**Conclusion**

A double ALNT seems to be a promising effective treatment for primary lower-limb lymphoedema, with a satisfactory reduction in limb perimetry, improvement in skin quality and high patient satisfaction.

**Conflict of interest**

None.

**Funding**

None.
References


