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Haemodynamic and Clinical Assessment of Lateral Marginal Vein Excision in Patients with a Predominantly Venous Malformation of the Lower Extremity

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Objective. *The purpose of the present study was to determine the effects of the surgical excision of lateral marginal veins (LMVs) in patients with a venous malformation (VM) affecting the lower extremity.*
Methods. *Preoperative and postoperative air plethysmography (APG), CEAP classification C scores, and venous clinical severity scores (VCSS) of the 25 VM patients who underwent LMV excision were compared.*
Results. *After LMV excision, venous haemodynamic parameters revealed significantly increased ejection fraction (EF, 33.2 S.D.18.5% vs. 39.7 S.D.21.2%, P = .020), and reduced venous volume (VV, 235.0 S.D.141.8 ml vs. 198.0 S.D.114.1 ml, P = .016) and residual venous fraction (RVF, 62.4 S.D. 26.6% vs. 56.9 S.D. 25.3%, P = .046). Clinical assessments of affected limbs revealed significantly improved mean CEAP C scores and VCSS (preoperative score, 4.4 S.D.1.7 vs. post-operative score 2.4 S.D.1.7, P = .026) after LMV excision versus preoperative data.*
Conclusion. *Haemodynamic and clinical improvements were observed in patients with lower extremity VM after LMV excision.*

Keywords: Vascular malformation; Lateral marginal vein; Assessment of outcome.

Introduction

Congenital vascular malformations (CVMs) can be classified as low-flow or high-flow. Low-flow malformations include venous, capillary, lymphatic, and combined form malformations, whereas high-flow malformations include arterial and arteriovenous malformations.[1] Venous malformations (VMs) commonly occur combined with capillary and/or lymphatic malformations (LMs). The presence of lateral marginal vein (LMV) is not uncommon in patients with a lower extremity VM. They are characterized by a greater diameter than normal superficial veins and a superficial location along the lateral aspect of the lower extremity. They are also referred to as marginal veins, lateral embryonal veins, or as a lateral venous anomaly. LMVs are often associated with deep venous anomalies such as aplasia or hypoplasia. In patients with deep venous aplasia of the lower extremity, LMV provides collateral venous channel for the affected limb with other venous collaterals, such as, an enlarged great saphenous vein or a persistent sciatic vein.[2]

LMV provide a source of venous reflux due to the absence of a venous valve, which can eventually cause persistent venous hypertension of the lower extremity. Though the precise mechanism is not known, chronic venous hypertension during childhood has been suggested as a cause of limb length discrepancy. Various surgical procedures[2,3] and embolisation procedures[4,5] have been used to remove abnormal vessels and to correct venous hypertension in patients with lower extremity VM. However, we were unable to find a report concerning the hemodynamic effects of LMV excision in patients with VM. The aim of the present study was to determine haemodynamic and clinical outcomes after LMV excision in patients with a lower extremity VM.

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Patients and Methods

Diagnosis and patient selection: Between May 1996 to October 2004, 580 patients with a VM (pure VM, 472 (81%) and combined type VM, 108 (19%)) registered at the CVM clinic of the Samsung Medical Center, Seoul. An analysis of the anatomic distribution of these VM lesions revealed that lower limbs and buttocks were the sites most frequently affected (Table 1).

To evaluate VMs, we performed whole body blood pool scans (WBBPS) with radionuclide (^{99m}Tc labelled RBC), MRI (Sigma horizon 1.5T, GE Medical, Milwaukee, WI, USA), long bone spot scanography, duplex ultrasonography (Ultramark9, ATL Inc, Bothell, WA, USA) of the affected limb, and radionuclide (Tc-99m antimony sulphide colloid) lymphoscintigraphy if indicated. The timings of these examinations depended on patient age, clinical features, and compliance.

Among the 274 patients with a lower extremity VM, 98 (35.8%) patients had LMV by inspection or by the above-described imaging studies. Of those with an LMV, the frequency of an associated deep venous anomaly such as aplasia or hypoplasia (diameter of hypoplastic segment of deep vein $<$ 50% of adjacent vein diameter) was 13.3% (Fig. 1). We performed LMV excisions in 25 limbs (right leg 9, left leg 16) of 25 patients (15 males, 10 females). Our indications for LMV excision were prominent LMV accompanied with clinical symptoms of chronic venous insufficiency (CVI), thrombus in LMV, or significant limb length discrepancy ($>$2 cm) in paediatric patients (Fig. 2). When LMV was associated with the deep venous agenesis or hypoplasia of the ipsilateral leg, we regarded it as a contraindication for LMV excision. We also excluded the patients from the candidate for LMV excision when LMV was not prominent or the clinical manifestation of CVI was absent or minimal.

Table 1. Anatomic distribution of venous malformations (VMs)

Distribution	Type of Vascular Malformation		Total (%)
	Pure VM	Combined VM with LM and/or CM	
Head and neck	120	32	152 (26.2)
Trunk:	37	3	40 (6.9)
Chest	20	2	22 (3.8)
Abdomen and pelvis	11	1	12 (2.1)
Perineum, genitalia	6	0	6 (1.0)
Extremity:	285	60	345 (59.5)
UE	60	11	71 (12.2)
LE and/or buttock	225	49	274 (47.2)
Multi-focal	30	13	43 (7.4)
Total	472	108	580 (100)

VM, venous malformation; LM, lymphatic malformation; CM, capillary malformation; UE, upper extremity; LE, lower extremity.

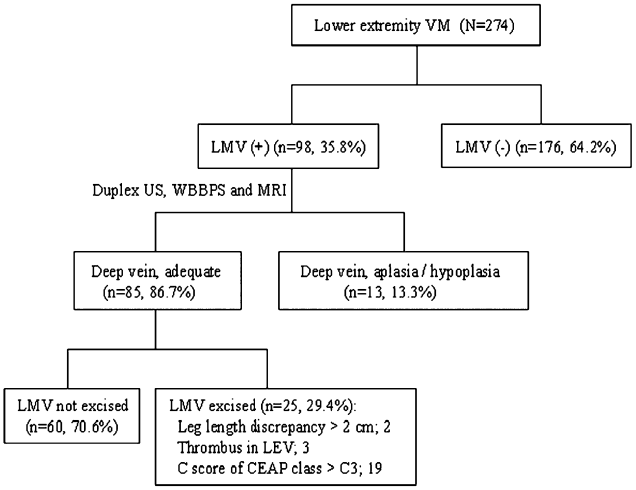

Fig. 1. Selection of candidates for lateral marginal vein (LMV) excisions among patients with a lower extremity venous malformation (VM).

Table 2 demonstrates the clinical features of the 25 patients who underwent LMV excision. One patient with an LMV thrombus developed symptomatic pulmonary embolism (PE) and an active venous ulcer before LMV excision.

Preoperative preparation and operation: To predict the haemodynamic outcome of LMV excision, we performed an LMV compression test preoperatively. The test was designed to observe the adaptability of the deep venous system after LMV removal. Using duplex ultrasonography, changes in maximal diameter and deep vein flow velocity were measured whilst manually compressing LMVs.

Before LMV excision, venous mapping was performed by ultrasonography. For patients with a potential risk of a massive haemorrhage, such as, those with a huge LMV, a sterile proximal thigh pneumatic cuff and cell saver were prepared before making a skin incision.

For LMV excision, the LMV was exposed through one or 2 longitudinal skin incisions under general anesthesia. After exposing the LMV, ligation and division of side branches, and secure suture closures of its distal and proximal ends were performed before removal.

In 18 patients, LMVs were excised in one stage (13 calf LMVs and 5 calf and thigh LMVs), whereas in 7 patients (28%) they were excised using staged operations (calf LMV excision first and thigh LMV excision later). The decision whether to adopt a one-stage or a two-stage operation was based on anatomical

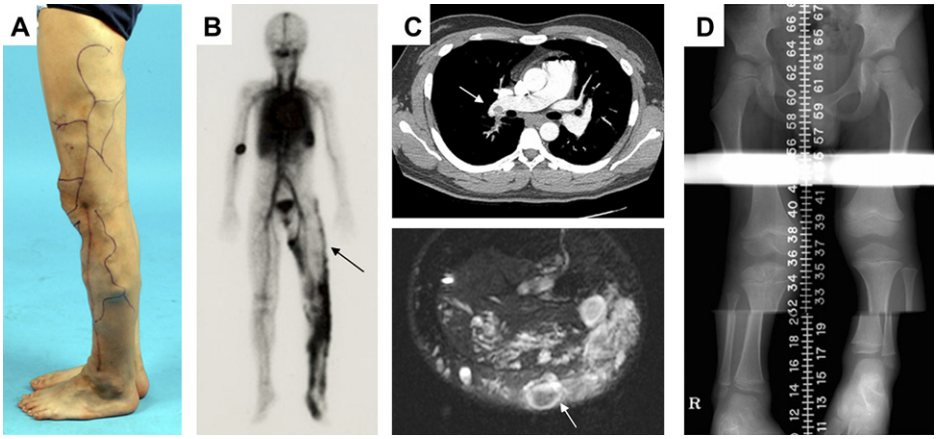

Fig. 2. Candidates for lateral marginal vein (LMV) excisions among patients with venous malformation; A, Prominent LMV on the left leg: venous mapping before LMV excision; B, A whole body blood pool scan (WBBPS) revealed an LMV (arrow) and a venous malformation (VM) in the left leg; C, Top: spiral CT scan revealing a pulmonary embolism (arrow) Bottom: MRI shows a venous malformation in lower extremity muscle and an LMV thrombus (arrow) in a same patient; D, Scanogram of the lower extremity revealing a bone length discrepancy in a VM patient.

features of the LMV and the patient's ability to tolerate the operation. One of the reasons why we perform LMV excision as a staged operation was to avoid large amounts of intraoperative bleeding, particularly in paediatric patients. Another reason was to reduce the sudden haemodynamic impact of LMV excision on the deep venous system, as we expected patient tolerance to increase to the new haemodynamic environment after partial excision.

Following LMV excision, we provided bed rest with leg elevation for a minimum of 24 hours, and then initiated ambulation with an elastic compression stocking as tolerated; generally longer bed rest was required for haemostasis than for varicose stripping. Low dose unfractionated heparin or prophylactic low molecular weight heparin was given for a week.

Table 2. Preoperative demographic and clinical data of 25 patients that underwent lateral marginal vein (LMV) excisions

Data	$N = 25$		
Age (y), Mean ± SD (Range)	17.2 ± 12.6 (4–53)		
Gender (Male : Female)	15 : 10		
Involved limb (Right : Left)	9 : 16		
Limb length discrepancy on scanogram ($n = 25$)	Shorter	Longer	Subtotal
≥2 cm	1	1	2 (8%)
1–2 cm	0	6	6 (24%)
<1 cm	3	14	17 (68%)
Lymphoscintigraphic findings ($n = 14$)			
Main lymphatic channel obstruction	3/14 (21.4%)		
Diminished lymphatic clearance with patent main lymphatic channel	11/14 (78.6%)		

Assessment of surgical results: To evaluate the haemodynamic effects of LMV excision, we compared preoperative and postoperative APG findings 1 month post-operatively. APG (ACI Medical, Sun Valley, CA) was performed according to the protocol described by Christopoulos *et al.*[6,7] by registered vascular technicians. Comparative APG data was available in 13 patients (7 calf LMV excisions, 6 calf and thigh LMV excisions), but unavailable in 12 paediatric patients due to poor cooperation or the lack of appropriately sized cuffs.

We also assessed the clinical outcomes of LMV excisions in all patients by comparing pre- and postoperative C scores according to the CEAP classification[8] and venous clinical severity scores (VCSS).[9] Postoperative clinical assessments were performed at 25 S.D. 21 months (range 2–74 months) after LMV excisions.

Statistical method: Data are presented as means and S.D. and statistical significance was determined using the paired t-test. P values of <0.05 were considered statistically significant.

Results

Preoperative ultrasonography of LMVs revealed venous reflux in all patients and thrombus in 3 (12%). LMVs were distributed over the whole leg in 88% of patients, and drained into the femoral vein in 84% of patients (Table 3). A preoperative LMV compression test revealed increased diameter and flow velocity in deep veins during LMV compression (Table 4).

Table 3. Location and morphologic features of lateral marginal veins (LMVs) by duplex ultrasonography and MRI

Features of LMV	$N = 25$
Location	
Calf only	3 (12%)
Calf and thigh	14 (56%)
Calf, thigh, and buttock	8 (32%)
LMV drain into:	
Popliteal vein	2 (8%)
Femoral vein	21 (84%)
Great saphenous vein	1 (4%)
Short saphenous vein	1 (4%)
Thrombus in LMV	3 (12%)

Table 5. Changes in air plethysmography (APG) parameters after lateral marginal vein (LMV) excision ($n = 13$ limbs)

APG parameter	Mean ± SD		*P* value[a]
	Preoperative	Postoperative	
Venous volume (ml)	235.0 ± 141.8	198.0 ± 114.1	.016
Venous filling index (ml/sec)	5.3 ± 6.4	3.2 ± 2.6	.063
Ejection volume (ml)	68.9 ± 58.8	67.9 ± 41.5	.886
Ejection fraction (%)	33.2 ± 18.5	39.7 ± 21.2	.020
Residual venous fraction (%)	62.4 ± 26.6	56.9 ± 25.3	.046

[a] Paired t-test.

After LMV excisions, APG revealed a significantly reduced postoperative venous volume (ml) and residual venous fraction (%) and an increased ejection fraction (%) (Table 5). Clinical assessment using CEAP classification C score and VCSS revealed an improved clinical status after LMV excisions (Table 6).

No operative mortality or major morbidity occurred during the early postoperative period, except for lymphatic leakage from the wound in 3 patients. All lymphatic leakages occurred in patients with a coexisting lymphatic malformation. As a late complication, recurrent lymphangitis and cellulitis developed in 2 patients (8%) on the operated limb.

Discussion

Klippel-Trenaunay syndrome (KTS) is a relatively well-known type of CVM that is composed of venous, lymphatic and capillary malformations, and which is associated with soft tissue or bony limb hypertrophy.[10] Two types of surgical treatment have been attempted in patients with VM: reconstruction of abnormal deep veins and ablation of abnormal veins.

Table 4. Results of preoperative lateral marginal vein (LMV) compression test[a]

Deep vein examined	Measured value (mean ± SD)		Mean Augmentation (%)
	Before LMV Compression	During LMV Compression	
Femoral vein:			
Diameter (mm)	5.4 ± 2.9	6.0 ± 3.3	13.8 ± 10.1
Velocity (cm/s)	12.9 ± 4.3	21.1 ± 9.9	61.6 ± 41.3
Popliteal vein:			
Diameter (mm)	5.3 ± 2.5	5.9 ± 2.7	14.8 ± 8.3
Velocity (cm/s)	11.1 ± 6.9	16.5 ± 8.8	8.4 ± 39.3

[a] LMV compression test indicates the measurement of deep venous diameter and flow velocity by duplex ultrasonography before and during manual compression of the lateral marginal vein (LMV) in the supine position.

For the reconstruction of the deep venous system, the release of an extrinsic compressive band,[2] competent vein transposition,[11] and deep venous reconstruction using a contralateral saphenous vein graft[12] have been reported. Ablation procedures include the surgical excisions of incompetent veins, such as, LMVs, saphenous veins or varicosities[13] and embolo-sclerotherapy for abnormal venous clusters.[5]

We use embolo-sclerotherapy to treat diffuse infiltrating VMs, though not for LMVs. There is a significant risk of skin damage if a strong sclerosant is used to treat a superficially located LMV. Though Jacob *et al.*[14] reported LMV in 72% of KTS, we found LMV in 36% of lower extremity VM patients, which included KTS and pure VM patients. Before excising an LMV, an assessment of the deep venous system is mandatory to avoid worsening venous hypertension caused by ablating an LMV in a patient with deep venous hypoplasia or aplasia.

During LMV compressions above and below the knee, we observed increased diameter and flow

Table 6. Clinical outcomes after lateral marginal vein (LMV) excisions

Clinical outcomes	No. of limb (%)		*P* value
	Preoperative	Postoperative	
C score[a]			.001[c]
C_1	0	8 (32%)	
C_2	6 (24%)	1 (4%)	
C_3	15 (60%)	13 (52%),	
C_4	3 (12%)	2 (8%)	
C_5	0	1 (4%)	
C_6	1 (4%)	0	
VCSS[b]	4.4 ± 1.7	2.4 ± 1.7	.026[d]

[a] C score according to CEAP classification. C1, telangiectases, reticular veins, malleolar flare; C2, varicose veins; C3, edema without skin changes; C4, skin changes ascribed to venous disease (e.g. pigmentation, venous eczema, lipodermatosclerosis); C5, skin changes as defined above with healed ulceration; C6, skin changes defined above with active ulceration.
[b] VCSS: venous clinical severity score.
[c] GEE, Generalized Estimating Equations.
[d] Paired t-test.

velocity at the femoral and popliteal veins in patients with non-stenotic deep venous system. In patients with deep venous hypoplasia, we observed diameter increase in the hypoplastic segment was minimal but the venous flow velocity increased. This test provides physiological evidence of deep venous congestion due to a sudden increase in blood flow into the deep venous system after LMV excision. Though this test requires further assessment to determine it value in predicting venous gangrene, it can be used with discretion as a part of an overall assessment to reduce the risk of sudden venous congestion in patients with deep venous aplasia or hypoplasia.

Eifert *et al.*[15] reported that 47% of VM patients have various types of deep venous anomalies, but deep venous aplasia or hypoplasia were reported to account for only 8% of these anomalies. We found deep venous aplasia or hypoplasia in 13% of patients with an LMV. According to Servelle's extensive review[3] of surgical KTS treatment, deep venous anomaly was identified most often at the popliteal vein (51%), followed by the femoral vein (16%), both femoral and popliteal veins (29%), iliac vein (3.3%), and the inferior vena cava (0.7%). In addition, they reported that venous aplasia was more common at the femoral segment.

It is difficult to determine the effects of LMV excision from previous reports, because of the range of surgical procedures that were used.[2,13] A recent series[6] reported clinical improvement but a high recurrence rate for varicosities after treatment. However, their indications for surgical treatment differed from ours. In particular, we observed LMV thrombus in 3 patients (12%). In one of these 3 patients, symptomatic PE coexisted at presentation although this has only rarely been reported.[16] During LMV excision, intraoperative haemorrhage is the main operative risk because bleeding from a primitive vessel is difficult to control using standard vascular techniques. In our series, 3 patients (12%) required a blood transfusion of more than 5 units during LMV excision. We recommend that pneumatic tourniquet compression of the leg and the use of a cell saver machine when massive bleeding occurs during LMV excision.

Leg length discrepancy is another important issue in patients with a CVM. Some previous reports have suggested that early, active vascular interventions used to correct venous hypertension can prevent lower limb length discrepancy in paediatric VM patients.[17,18] The effect of LMV excision on bone growth has not been determined but we recommend LMV excision before the completion of long bone growth or the development of the advanced signs of CVI. The mean age of our patients was 17.2 S.D. 12.6 years in the present series.

Lymphatic anomalies may not be clinically apparent until a later in life in cases of combined type veno-lymphatic malformations. Radionuclide lymphoscintigraphy showed main lymphatic channel obstruction while others showed diminished lymphatic clearance without major channel obstruction. We experienced 2 patients who developed worsening late limb swelling due to recurrent lymphangitis and cellulitis following LMV excision. In these 2 patients, a preoperative lymphoscintigram showed diminished lymphatic clearance without major lymphatic obstruction in the affected limb, and follow-up lymphoscintigram showed further diminution of lymphatic clearance after LMV excision.

For patients with lymphangitis, CEAP classification C scores did not reflect worsening of limb swelling, though a comparison of pre- and postoperative C-scores showed a statistically significant improvement. In the present study, we found that CEAP classification C scores are inappropriate for outcome assessment in VM patients.

Other reports have described the role of APG in the haemodynamic assessment of CVI,[19,20] and in the postoperative evaluation of superficial vein ablation in CVI patients.[21,22] Although we accept that APG cannot provide complex haemodynamic information in VM patients, we observed that venous haemodynamics were significantly improved after LMV excision.

Two major complications of LMV excision are intraoperative haemorrhage and the late occurrence of lymphangitis and cellulitis. To avoid the operative risk of haemorrhage, we believe that endovascular LMV ablation may become a future alternative option. However, preoperative lymphoscintigraphic findings of a patent main lymphatic channel cannot guarantee freedom from later lymphatic complications after LEV excision.

We recommend LMV excision in patients with a prominent LMV with clinical manifestations of CVI or of thrombus in an LMV. Even though we do not provide evidence of the beneficial effect of LMV excision in paediatric patients with a leg length discrepancy, the long-term outcome of its effect on bone growth should be followed in this patient group. Before LMV excision is considered, anatomical and functional assessments of the deep venous system with imaging studies and LMV compression ultrasonography help in the selection of suitable patients.

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