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原　著

婦人科系疾患に続発した下肢リンパ浮腫例のリンパシンチによる検討

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Lymphoscintigraphy of the patients with lymphedema after gynecologic operations

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Abstract

Lymphedema after gynecologic operations, particularly gynecologic cancer treatments, is one of common complications. Lymphedema, once developed, becomes chronic and refractory. The purpose of this study is to evaluate the lymphatic functions in lymphedema developed after gynecologic operations with lymphoscintigraphy.

Images of lymphoscintigraphy in 100 patients were classified into five types. Majority of the images in the bilateral limbs of the bilateral lymphedema could be classified into type I. Majority of the images in the affected limbs of the unilateral lymphedema could be classified into type IV. Majority of the images in the non-affected limbs of the unilateral lymphedema could be classified into type I. About 60% of the lymphoscintigraphic images of the non-affected side had abnormal findings. Seventeen of the 100 cases underwent lymphoscintigraphy twice. Ten of the 17 cases changed them types of lymphoscintigraphic images.

Lymphoscintigraphy is useful method to evaluate functions and it is suspected that there may be some possibilities in patients with unilateral lymphedema after gynecologic operations to develop lymphedema in contralateral limb.

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Key words : lymphedema, lymphoscintigraphy, gynecologic operation, lower limb, lymphatic function

はじめに

婦人科領域疾患，特に骨盤内リンパ節郭清を伴う悪性腫瘍術後の合併症の一つに下肢リンパ浮腫が挙げられる．発症すると難治性であり，その予防は極めて重要である．婦人科領域術後の下肢リンパ浮腫について，発症要因や頻度についての報告は散見されるが[1~10]，発症後のリンパ機能について評価した報告は文献を渉猟し得た限りない．

今回，婦人科領域疾患術後にリンパ浮腫を発症し，リンパ浮腫の治療目的に当科を受診した患者にリンパシンチグラフィー（以下リンパシンチ）を施行した．得られた画像を分析・検討し，さらに浮腫の経時的変化により複数回リンパシンチを施行した症例において画像の変化を分析したので報告する．

対象と方法

1992年5月～2008年12月までに婦人科領域の疾患で手術を施行し，術後に生じた下肢の浮腫を主訴に当科を受診した100例を対象とした．年齢は30～86歳，平均62±12.3歳，原疾患は子宮癌92例，卵巣癌6例，子宮筋腫と子宮内膜症がそれぞれ1例であった．所属リンパ節郭清を施行したのは81例，施行していないのは3例，郭清についての詳細が不明であるのは16例であった．

術後放射線治療を施行したのは44例，施行していないのは47例，詳細が不明であるものは9例であった．

100例（200肢）にリンパシンチを施行した．得られた画像を鮑ら[12]，Maegawaら[13]が分類する５つのタイプに分類し，下肢のリンパ機能を評価した．100例のうち17例（34肢）では，リンパシンチを２回施行し，画像の変化を比較した．また同時に臨床的重症度の評価として，下肢の周径を足中央部，足関節上，下腿（膝蓋骨下10cm），大腿（膝蓋骨上10cm）で測定し，片側浮腫症例では両下肢を比較し，検討した．

片側浮腫症例における左右の発症頻度についてカイ二乗検定を用いて検討を行った．また全症例においてリンパ浮腫の発症における放射線治療の関連についてカイ二乗検定を用いて検討を行った．また，Maegawaらによると，リンパシンチのタイプ分類は重症度と相関するとしており[13]，放射線治療によるリンパシンチタイプ分類の違いについてマンホイットニー順位和検定を用いて検討した．$P<0.05$を統計学的に有意差があるとした．

リンパシンチは両足背の皮下に99mTc標識ヒトアルブミンを注射し，その30分後と120分後にシンチカメラで撮影した．

タイプⅠはトレーサー注入から30分後の撮影で鼠径リンパ節が描出され，また，注入部から鼠径にかけ，大伏在静脈に沿い集合リンパ管が描出されることが多い．正常のリンパシンチ所見もタイプⅠに分類される．タイプⅡは大腿部のみにDermal backflow（以下DBF）を認め，鼠径部より中枢側のリンパ通過障害により，それより末梢の大腿でのリンパ管内圧が高まって皮下に逆流している．タイプⅢは大腿と下腿にDBFを認め，タイプⅡよりさらに下腿でもリンパ管内圧が高まり大腿と下腿でリンパが皮下へ逆流している．タイプⅣは下腿のみにDBFを認め，大腿でのリンパ管機能は失われている，あるいは浮腫のために組織圧が著しく高まり，リンパがほとんど流れない状態である．タイプⅤは足，足関節周囲のみにDBFを認め，リンパ管機能は大腿・下腿ともに失われている．

結　果

・初診時の片側性と左右差について

両側性29例（29%），片側性71例（71%）であった．片側性のうち，右側下肢は27例（38%），左側下肢は44例（62%）であり，左右の発症頻度に有意差を認めた．（$P=0.044$）．

・片側例における周径による重症度分類

最大となる健側との周径差が０～２cmの軽症11例（15.5%），２～６cmの中等症24例（33.8%），６cm以上の重症35例（49.2%），不明２例であった．

・リンパシンチにおけるタイプ分類

両側例（29例，58肢）では，タイプⅠが26肢，タイプⅡが７肢，タイプⅢが７肢，タイプⅣが10肢，タイプⅤが８肢であった（Fig. 1）．

片側例（71例）の患側下肢（71肢）は，タイプⅠが８肢，タイプⅡが12肢，タイプⅢが16肢，タイプⅣが24肢，タイプⅤが９肢であった．健側下肢（71肢）はタイプⅠが67肢，タイプⅡが２肢，タイプⅢが２肢，タイプⅣとタイプⅤは認めなかった（Fig. 2）．健側下肢のタイプⅠを示した67肢を更に検討すると，リンパ排泄の遅延，鼠径リンパ節の減少，リンパ管の拡張や側副路の発達などのリンパ機能の異常を示す所見（Fig. 3－7）を41肢（61.2%）で認めた．（Table 1）

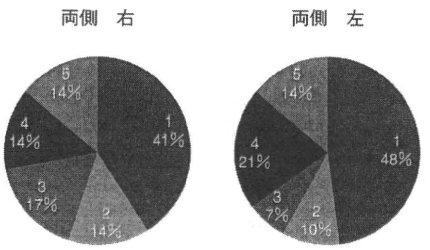


Fig. 1　Types of lymphoscintigraphic images in the bilateral lymphedema
Majority of the images in the bilateral limbs of the bilateral lymphedema could be classified into type I.

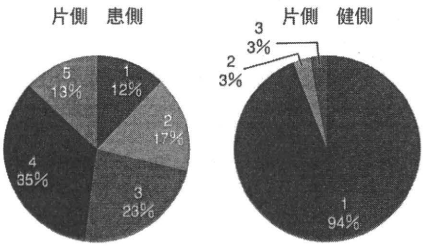
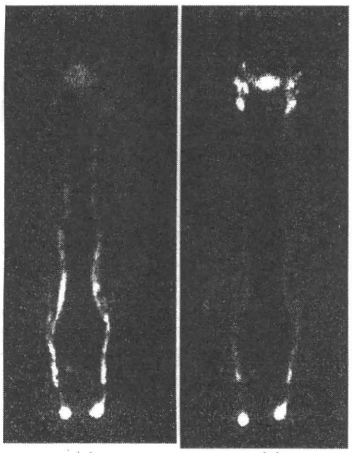


Fig. 2　Types of lymphoscintigraphic images in the unilateral lymphedema
Majority of the images in the affected limbs of the unilateral lymphedema could be classified into type IV.
Majority of the images in the non-affected limbs of the unilateral lymphedema could be classified into type I.

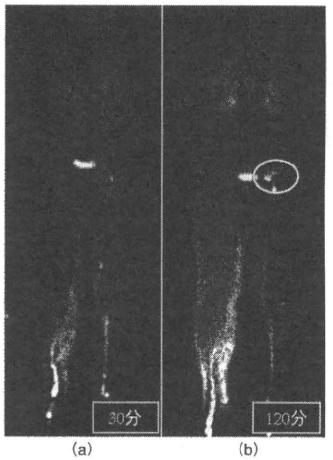


(a) (b)

Fig. 3 Normal lymphoscintigraphic images
(a) 30minutes after injection
(b) 120minutes after injection
Lymph collecting vessel and inguinal lymph node were observed in the image of 30 minutes after injection (a). In the image of 120 minutes after injection (b), lighter lymph collecting vessel was observed than 30 minutes after injection.

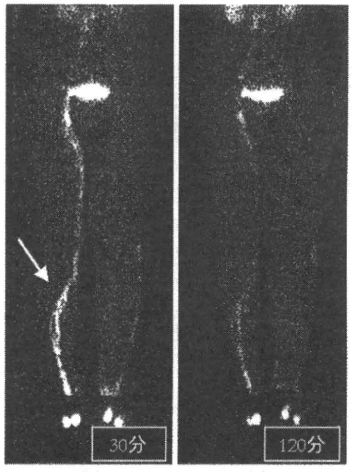
(a) (b)

Fig. 4 Delay in lymph drainage
(a) 30minutes after injection
(b) 120minutes after injection
Lymphoscintigraphic images of right lower limb lymphedema: Thick lymphoduct was observed in the image of 120 minutes after injection.(b)



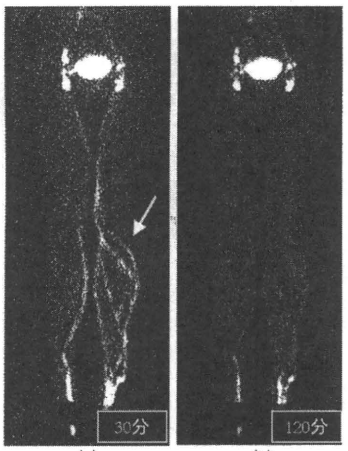
(a) (b)

Fig. 5 Decrease in the number of the inguinal lymph node
(a) 30minutes after injection
(b) 120minutes after injection
Lymphoscintigraphic images of left lower limb lymphedema: The number of the inguinal lymph nodes decreased. (circle)



(a) (b)

Fig. 6 Dilated lymphoduct
(a) 30minutes after injection
(b) 120minutes after injection
Lymphoscintigraphic images of right lower limb lymphedema: dilated lymphoducts were observed. (arrow)



(a) (b)

Fig. 7 Bypass of lymphoducts
(a) 30minutes after injection
(b) 120minutes after injection
Lymphoscintigraphic images of left lower limb lymphedema: bypass was observed. (arrow)

Table 1 Abnormal findings in the lymphoscintigraphic images of the non-affected limbs classified into type I

異常所見	排泄遅延	リンパ管拡張	リンパ節減少	側副路
肢	22／41 (53.4%)	20／41 (48.8%)	9／41 (22.0%)	16／41 (39.0%)

※所見は重複あり

・放射線治療について

両側例の29例中14例，片側例の71例中30例で放射線治療を施行しており，両側例の29例中11例，片側例の71例中34例で放射線治療を施行していなかった．リンパ浮腫の発症における放射線治療の関連は統計学的に有意差を認めなかった．(P＝0.753) また，放射線治療によるリンパシンチタイプ分類の違いについても統計学的な有意差を認めなかった．(P＝0.27) (Fig. 8)

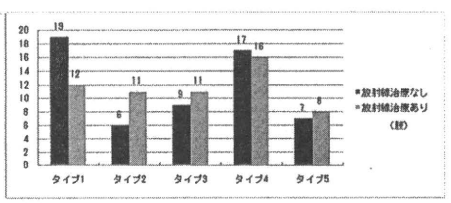


Fig. 8 Types of lymphoscintigraphic images according to radiation therapy
There is no significant difference in types of lymphoscintigraphic images according to radiation therapy.

・リンパシンチの経時的変化

リンパシンチを2回施行した症例は17例34肢であり，1回目のリンパシンチの撮影から平均25±10.7ヵ月後に2回目の撮影を施行した．17例中3例は理学療法のみ，他14例は手術（リンパ管静脈吻合術）施行例であった．両側例が1例，片側例が16例（右側6例，左側10例）であった．両側例の1例は理学療法のみで，リンパシンチのタイプ分類に変化がなかった．

片側例の患側では，タイプの改善を認めたものが5肢，不変が7肢，悪化が4肢であった．理学療法のみが2例であり，1例でタイプが悪化，1例で改善していた．片側例の健側では，初回のリンパシンチは1肢のみタイプⅢであり，他は全てタイプⅠであった．タイプが改善したものが1肢（1回目がタイプⅢであった症例），不変が14肢，悪化が1肢であった．不変であった14肢はタイプⅠのままであったが，4肢で異常所見の増加を認めた．

リンパシンチの経時的変化における代表症例を呈示する．

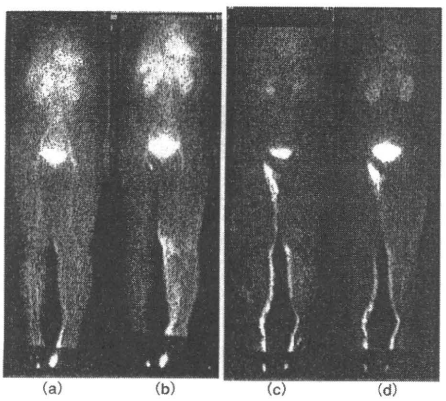
症例1：54歳　女性．片側例で健側のリンパシンチ所見が悪化した症例．

既往歴：36歳時に子宮体癌の診断で，準広範子宮全摘及び骨盤内リンパ節郭清術（放射線治療は不明）を施行した．

現病歴：術後15年頃より左下肢の浮腫が出現し，左下肢続発性リンパ浮腫の診断でバンテージ療法とマッサージ療法を施行するも改善せず，当科を紹介受診となった．

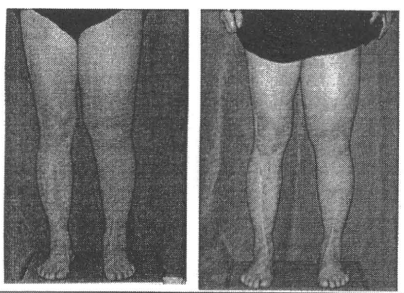
治療経過：初診時のリンパシンチは右タイプⅠ，左タイプⅣであった．バンテージ療法目的に入院治療を行い，その3ヵ月後に左下肢リンパ管静脈吻合術を施行（下腿で3吻合，大腿で1吻合，全て側端吻合）した．術後，下腿の浮腫は改善したが，左大腿の浮腫が残存していたため，2年後に再度，左下肢リンパ管静脈吻合術（下腿で2吻合，大腿で3吻合，全て側端吻合）を施行した．2回目の術前のリンパシンチでは，右がタイプⅡに悪化していた．2回目の術後，左下肢は浮腫の改善を認めるが，右下肢大腿のリンパシンチでDBFを認めた部位に他覚的に浮腫を認めた（Fig. 9, 10）．

症例2：58歳　女性．患側のリンパシンチ所見が悪化した症例．



(a) (b) (c) (d)

Fig. 9 Case 1: lymphoscintigraphic images
(a), (b): lymphoscintigraphic images at the first time (a) 30minutes after injection, (b) 120minutes after injection
Right lower limb was classified into type I, left lower limb was classified into type IV.
(c), (d): lymphoscintigraphic images 2 years after the first lymphoscintigraphy (before second operation) (c) 30minutes after injection, (d) 120minutes after injection
Right lower limb was classified into type II, left lower limb was classified into type IV. Dermal backflow on the right thigh was observed. Type of lymphoscintigraphic images changed from type I into type II on the right lower limb.



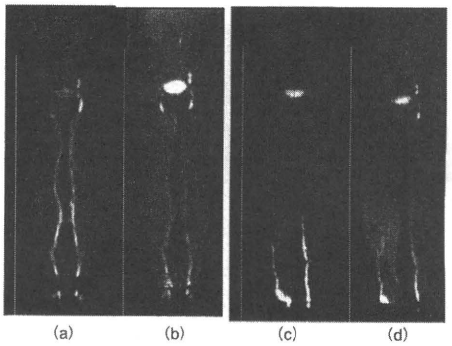
1回目術前 周径 (足背－足首－下腿－大腿)	2回目術後 周径 (足背－足首－下腿－大腿)
右(204－194－330－415)mm	右(224－192－336－449)mm
左(222－222－376－520)mm	左(219－198－345－516)mm

Fig. 10 Case 1: progress of treatment (at the first time and after second operation)
Lymphedema on the left lower limb improved. Lymphedema on the right thigh was observed.

既往歴：52歳時に子宮体癌の診断で，子宮全摘，リンパ節郭清を施行した．

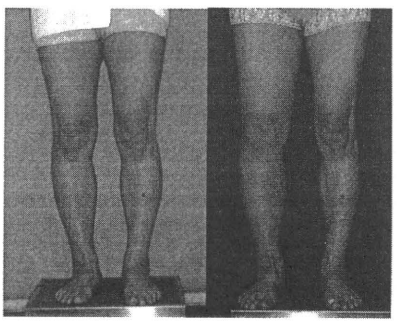
現病歴：子宮体癌術後から外陰部から右下肢にかけて浮腫を自覚していたが放置し，手術から2年後に当科を受診した．

治療経過：初診時のリンパシンチは右タイプⅡ，左タイプⅠであった．理学療法（マッサージ，ストッキング）を施行していたが，初診から3年後に浮腫の増悪を認めた．リンパシンチを再度施行したとこ



(a) (b) (c) (d)

Fig. 11 Case 2: lymphoscintigraphic images
(a), (b): lymphoscintigraphic images at the first time (a) 30minutes after injection, (b) 120minutes after injection
Right lower limb was classified into type II, left lower limb was classified into type I.
(c), (d): lymphoscintigraphic images 3 years after the first lymphoscintigraphy (c) 30minutes after injection, (d) 120minutes after injection
Right lower limb was classified into type IV, left lower limb was classified into type I. Dermal backflow on the right crus was observed. Type of lymphoscintigraphic images changed from type II into type IV on the right lower limb.



初診時 周径 (足背－足首－下腿－大腿)	3年後 周径 (足背－足首－下腿－大腿)
右(221－215－380－437)mm	右(236－220－392－462)mm
左(224－212－374－438)mm	左(230－202－348－392)mm

Fig. 12 Case 2: progress of treatment (at the first time and before operation)
Lymphedema on the right lower limb got worse especially on the right thigh.

ろ，右タイプⅣ，左タイプⅠと患側での所見の悪化を認めた（Fig.11，12）．右下肢に対し，リンパ管静脈吻合術（下腿で5吻合）を施行した．術後，浮腫の改善を認めた．

考　察

婦人科領域術後のリンパ浮腫発症頻度は諸家の報告では5～40%[2)~5)]といわれている．佐々木は第三次がん克服戦略事業の研究の結果，婦人科がん術後の下肢リンパ浮腫危険因子として高危険群を子宮癌の術後放射線療法，子宮癌の傍大動脈リンパ節郭清，中間危険群を卵巣癌の傍大動脈リンパ節郭清，子宮癌の高危険群以外，低危険群を卵巣癌の骨盤内リンパ節郭清のみと述べている[1)]．今回での検討では原疾患が子宮癌であった症例が全体の92%を占めていた．郭清の範囲は今回の検討に含めていないが，リンパ節郭清の有無については，郭清を施行していた患者は全体の81%であり，郭清を受けていないのは3例のみであった．放射線療法の浮腫に対する影響については多くの文献で発症が高まるとしている[1),2),7)~9)]が，発症に有意差を認めなかったとの報告もある[3),10),11)]．今回の検討は実際にリンパ浮腫が発症した症例の検討であるが，婦人科系疾患術後のリンパ浮腫症例では放射線治療の有無およびリンパシンチにおけるタイプ分類において統計学的な有意差を認めなかった．リンパシンチのタイプ分類は重症度と相関しており[13)]，重症化について放射線治療は関与していない可能性があると考えられる．

リンパシンチによるリンパ浮腫の診断・評価については多くの報告がある．多くの文献で異常所見として，鼠径リンパ節の描出が不良または欠損，DBFを挙げている[14)~19)]．Peckingら[14)]は，60分後の撮影における鼠径リンパ節の描出でリンパ疾患を持つか否かを診断できるとし，Yuanら[16)]は鼠径リンパ節描出の有無が重症度を示すとしている．その他，排泄の遅延，側副路の描出，リンパ管の拡張等が異常所見として挙げられている[14)~19)]．Yuanら[16)]は，側副路は正常でも認められるが，異常所見の一つとして挙げている．今回の検討では，側副路を認めることがリンパ浮腫の診断にはならないと考えるが，異常所見の一つとして挙げた．

リンパ浮腫の画像評価としては，ICGを用いた蛍光近赤外リンパ管造影がある．当科では，リンパ管静脈吻合術の際，リンパ管を同定する目的に使用している．今回の検討症例でも，手術を施行した症例では，蛍光近赤外リンパ管造影を行っており，リンパ管の描出およびDBFの描出はリンパシンチとほぼ同等の所見が得られた．しかし，蛍光近赤外リンパ管造影ではおおよそ皮膚から2cm以上深部の観察が不可能であり，大腿部ではシンチ上でリンパ管が同定されていても蛍光近赤外リンパ管造影では同定できないことが多かった．

リンパシンチの経時的変化についてPeckingらは，初回と複合的理学療法後で大きな変化を認めなかったとしており[14)]，Campisiらは，手術でのリンパ管吻合開存の確認にリンパシンチでの所見の変化を挙げている[20)]．2回のリンパシンチを施行した17例では，9例で患側肢のリンパシンチの所見の変化を認めた．治療の内容（理学療法，手術療法）に関わらず，リンパ機能の評価が変化していくことが示唆され，経時的にリンパ機能を評価することで，治療における評価と今後の臨床経過の予測が可能になると考えられた．

両側例と片側例を比較した文献は多くないが，光嶋らはリンパ浮腫の経過を一過性型（自然または保存的治療で治癒または軽快），慢性片側型（発症後浮腫が緩徐に増強するがあくまで片側に限局），慢性両側型（発症後浮腫が緩徐に増強するが中等症までの両側性），重症両側型（治療の有無に関わらず浮腫が増強し重症の浮腫になる．片側性から両側性になるもの）の4タイプに分類しており，下肢リンパ浮腫35例の内，慢性片側型を51%に認めたとしており，片側性から両側性に移行したものを，少なくとも5例に認めたとしている[21)]．今回の検討では両側例が29%であり，リンパシンチタイプ分類では左右ともタイプⅠが多かった．一方片側例は71%であり，患側はタイプⅣが多く，これに対し健側はタイプⅠが最も多く67肢（94.4%）であった．しかし，健側タイプⅠ症例でも41肢に何らかのリンパ機能の異常所見を認めており，タイプⅡ，Ⅲの症例も含めると片側例の健側は45肢（63.4%）にリンパ機能の異常所見が認められた．この異常所見は，リンパ機能が障害され，実際にはリンパ浮腫を発症しているが自覚がない軽症例とリンパ機能は障害されているがリンパ浮腫は発症していない不顕性例が含まれていることが示唆される．

また，経時的に片側例の健側肢において異常所見の増加を認めた症例は16肢中5肢（31.3%）であった．また，その中に他覚的に浮腫を認めた症例もあった．リンパシンチの異常所見の増加やタイプの重症化が浮腫を自覚する以前に認められており，所見の悪化が浮腫発症の予測になりうると考えられた．

片側例でも両側に移行する可能性があり，初診時の臨床所見のみで両側に移行するか否かを評価することは困難である．患側のみでなく，浮腫を認めていない健側肢に対してもリンパシンチによりリンパ機能の評価を行うことにより軽症例や不顕性例の発見を可能とし，また経時的に評価を行っていくことにより臨床所見や自覚症状のみでは評価困難なリンパ浮腫の進行を客観的に捉え，浮腫の経過を予測し，発症の予防に努めることが可能になると思われた．

ま と め

婦人科領域術後に生じた下肢リンパ浮腫に対し，リンパシンチを施行・検討した．片側例の健側肢71肢中45肢（63.4%）において，リンパシンチで何らかの異常所見を認め，経過観察が必要であると思われた．リンパシンチは患肢のリンパ機能評価のみでなく，片側例では健側肢の評価を行うことで身体所見や周径計測では評価困難なリンパ機能障害の進行度を捉え，経過予測・浮腫発症予防に効果的であると思われた．

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原発性リンパ浮腫12例の術前画像評価と術中所見の比較検討

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Comparison between preoperative lymphoscintigraphy and intraoperative double dye injection method in primary lymphedema

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Abstract

It is important that we detect functional lymphatics in lymphaticovenous anastomosis. We make a comparative review of preoperative lymphoscintigraphy and intraoperative double dye injection method in 12 cases of primary lymphedema.

The detection ratio of lymphatics in incision site per an affected limb is 70.7%.

In the incision site based on lymphoscintigraphy and intraoperative double dye injection method findings the ratio is 91.1%.

Out of a total of incision site,the detection ratio is 93.1% where we incised based on lymphoscintigraphy and intraoperative double dye injection method findings. The odds are low in the incision site beased on one findings only.

Compare the date from the point of view of incision site, The most common site where we incised based on lymphoscintigraphy only is thigh,the least site is dorsum of foot.

Lymphoscintigraphy is useful to detect functional lymphatics and to decide incision site preoperatively.

Key words： lymphoscintigraphy, primary lymphedema, lymphaticovenous anastomosis, indocyanine green

はじめに

われわれの施設ではリンパ浮腫の患者に対しリンパシンチグラフィーを施行し，診断とタイプ分類を行っている[1),2)]．しかしながら，原発性リンパ浮腫に対しては続発性リンパ浮腫と同じ分類にあてはまらない型が多い．リンパ管静脈吻合術の術前検査としてリンパシンチグラフィーを行い，リンパ管の走行をある程度知る事が可能であるが，原発性リンパ浮腫の場合に関して言及された報告はわれわれが渉猟し得た限りなかった．

今回，われわれは術前にリンパシンチグラフィーを施行し，術中二重色素造影法[3)]を用いてリンパ管静脈側端吻合術を行った原発性リンパ浮腫12例について検討したのでリンパシンチグラフィーと術中所見との関係について報告する．

対象と方法

2006年6月から2010年2月までに，下肢片側の浮腫を主訴に当院当科を受診し，リンパシンチグラフィーの結果，原発性リンパ浮腫と診断しリンパ管静脈側端吻合術を施行した12例（男性6例　女性6例）を対象とした．年齢は14歳から77歳までの平均47歳であり，早発性4例，遅発性8例であった．

リンパ管は二重色素造影法を用い，PDEカメラで経

皮的に皮下のリンパ管を同定した[4].

明らかな線状に観察出来た部位を実線，明らかな線状ではないがリンパ管の存在が示唆された部位を破線で体表にマーキングをした．これらをリンパシンチグラフィーと比較検討した．

症例提示

31歳男性，大腿部を中心とした左下腿浮腫を主訴に当院当科を受診した．

リンパシンチグラフィーでは左下腿内側を上行するリンパ管を認め，大腿内側にdermal back flow（以下DBF）を認めた（図１ａｂ）．

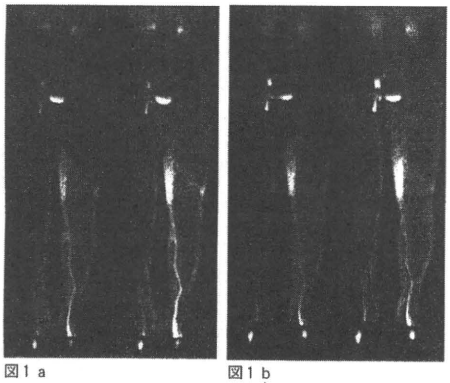

図１ａ　図１ｂ

Lymphoscintigraphic images of the lower limbs. a is 30min after injection of tracer. b is 60min after. Dermal backflow in the left thigh.We can see lymphatics in lower leg.

リンパ管を体表にマーキングし（図２ａ），これをリンパシンチグラフィー（図２ｂ）と比較した．大腿内側のDBFはリンパシンチグラフィーとPDEの両者で観察可能であった．足背から足関節前面を通り下腿中央から内側頭側へ向かうリンパ管も両者で確認できた．しかし，下腿前面を上行しDBFを形成するリンパ管はPDEカメラで確認できたが，リンパシンチグラフィーでは明らかではなかった．

手術は全例側端吻合術を施行し，顕微鏡下で開存を確認した．

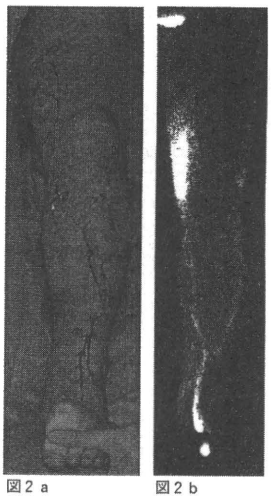

図２ａ　図２ｂ

Compare Intraoperative double dye injection method and lymphoscintigraphic image of the lower limb.

結　果

鮑らの報告[1]に従い，リンパシンチグラフィーの所見をリンパ管の集簇している部位で分類すると大腿型２例，下腿型６例，下腿・大腿型２例，低形成２例であった．

手術においては，切開部位においてリンパ管を同定できなかった部位も存在した．

リンパ管を同定できた割合を患肢あたりでみると，5.8切開中4.1切開（70.7%）であった．リンパシンチグラフィーと術中二重造影法の両方でリンパ管の存在が示唆されて切開した部位では患肢あたり1.9切開中1.8切開（91.1%）であり，リンパ管が同定可能であった割合は高い傾向にあった．

リンパ管が同定できた割合をリンパシンチグラフィーと術中二重造影法で比較すると，それぞれの検査でリンパ管の存在が示唆された部位においてはリンパ管が同定できた割合が高かった．リンパシンチグラフィーと術中二重造影法の両方でリンパ管の存在が示唆された切開部では，29切開中27切開（93.1%）でリンパ管が同定可能であり高い傾向にあった（表１）．

部位別に比較すると，リンパシンチグラフィーの所見のみに基づいて切開を行った部位は大腿部が多く，半数でリンパ管を同定できた．さらに，大腿部では術中二重造影法でリンパ管の存在が示唆された部位でも

表1　The detection ratio of lymphatics

リンパ管を同定できた確率		術中二重色素造影法		
		あり	なし	
リンパシンチグラフィー	あり	93.1% (27/29 切開)	50.0% (2/4 切開)	87.8% (29/33 切開)
	なし	82.1% (23/28 切開)	22.2% (2/9 切開)	67.6% (25/37 切開)
		87.7% (50/57 切開)	30.8% (4/13 切開)	

表2　The detection ratio of lymphatics per region

			術中二重色素造影法	
			あり	なし
大腿	リンパシンチグラフィー	あり	80% (4/5 切開)	50% (1/2 切開)
		なし	50% (2/4 切開)	50% (2/4 切開)
下腿	リンパシンチグラフィー	あり	93.8% (15/16 切開)	50% (1/2 切開)
		なし	94.1% (16/17 切開)	0% (0/2 切開)
足背	リンパシンチグラフィー	あり	100% (8/8 切開)	0% (0/0 切開)
		なし	71.4% (5/7 切開)	0% (0/3 切開)

リンパシンチグラフィーの所見の有無で差を認めた．これに対し，足背ではリンパシンチグラフィーの所見のみに基づいて切開を行った部位ではリンパ管を同定出来なかった（表2）．

考　察

リンパシンチグラフィーと術中二重造影法の両方でリンパ管の存在が示唆された部位ではリンパ管が同定可能である確率が高い傾向にあった．また，術中二重造影法でリンパ管の存在が示唆された部位でも，リンパシンチグラフィーの所見の有無でリンパ管を同定できる確率に差を認め，これは大腿部で顕著であった．大腿部の術中所見として，下腿や足背より深部にリンパ管を認めた事から，これはPDEカメラの感度が非常に高く，細い表在リンパ管を鋭敏に捕らえていることに加えて，リンパシンチグラフィーがPDEカメラでは観察出来ない深部のリンパ管も捕らえる事ができることが一因であると考えられる．

また，足背部において，重度の低形成な症例や足背でDBFが広がる症例ではリンパシンチグラフィーでも二重造影法でもリンパ管は確認できず，リンパ管静脈側端吻合術の適応を検討する必要があると思われた．

原発性リンパ浮腫は二次性と異なり，全体または局所のリンパ管が障害されていると考えられるが，リンパシンチグラフィーで描出されたリンパ管は同定可能なリンパ管と考えられ，リンパシンチグラフィーが術前検査として，特に大腿部・下腿部で有用であると考えられた．

ただし，術後の経過は症例によって様々であり，今後さらなる検討の必要がある．

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TYPES OF LYMPHOSCINTIGRAPHY AND INDICATIONS FOR LYMPHATICOVENOUS ANASTOMOSIS

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Several authors have reported the usefulness and benefits of lymphoscintigraphy. However, it is insufficient to indicate microvascular treatment based on lymphedema. Here, we present the relationships between lymphoscintigraphic types and indications for lymphatic microsurgery. Preoperative lymphoscintigraphy was performed in 142 limbs with secondary lymphedema of the lower extremity. The images obtained were classified into five types. Type I: Visible inguinal lymph nodes, lymphatics along the saphenous vein and/or collateral lymphatics. Type II: Dermal backflow in the thigh and stasis of an isotopic material in the lymphatics. Type III: Dermal backflow in the thigh and leg. Type IV: Dermal backflow in the leg. Type V: Radiolabeled colloid remaining in the foot. Lymphaticovenous anastomosis was performed in 35 limbs. The average number of anastomoses per limb was 3.3 in type II, 4.4 in type III, 3.6 in type IV, and 3 in type V. The highest number of anastomosis was performed in type III. In conclusion, type III is suggested to be the best indication for anastomosis compared with types IV and V. © 2010 Wiley-Liss, Inc. Microsurgery 30:437–442, 2010.

Lymphoscintigraphy has been widely adopted for the evaluation of lymphedema. Several authors[1–5] have reported its usefulness and benefits as a less invasive method of examination and described patterns of lymphoscintigraphy in patients with peripheral lymphedema. Establishment of a staging of lymphoscintigraphy was also tried for evaluating severity in lymph edema[4,5]. However, these patterns or staging is inadequate for surgeons to treat lymphedema by microvascular techniques, particularly for surgical indications. For the purpose of determining indications for microsurgery, we applied preoperative lymphoscintigraphy to patients with peripheral lymphedema and divided the obtained lymphoscintigraphic images into five types, in particular with an emphasis on patients with secondary lymphedema of a lower extremity. In this study, we present the relationships between lymphoscintigraphic types and clinical stages of lymphedema and between the types and indications for lymphaticovenous anastomosis.

MATERIALS AND METHODS

Between January 1999 and July 2008, lymphoscintigraphy was performed on 142 patients with lymphedema in a lower extremity. Of the 142 patients, 111 were diagnosed with secondary lymphedema and the other 31 were diagnosed with primary lymphedema or other disorders, which were excluded in this study. Of the included patients, 102 were female and nine were male. The average age of the 111 patients was 63 years, ranging from 38 to 92 years. The original disease of the secondary lymphedema was uterine cancer in 82 patients, colorectal cancer in eight, ovarian cancer in seven, prostate cancer in four, malignant lymphoma in three, and miscellaneous disorders in 10. Of the 111 patients, 80 had unilateral lymphedema (group A) and 31 had bilateral lymphedema (group B). Combined physical therapy was applied to the patients diagnosed as having secondary lymphedema before and after surgery.

A small amount (0.2 ml, 40 MBq) of technetium-99m-labeled human serum albumin was injected subcutaneously at the first and third web spaces of each foot, and passive muscular exercise was standardized to reduce the variability of lymphatic function. A gamma camera was used to obtain images of the limbs 30 and 120 minutes after injection. In our previous studies, inguinal lymph nodes and collecting lymphatics along the great saphenous vein can be detected 30 minutes after injection in the lower extremities without lymphatic problems. However, in some severe cases, the injected contrast medium was slowly transported proximally in the affected limb even 120 minutes after injection. Therefore, 30 and 120 minutes seemed to be good times for imaging for the mild cases and for the moderate or severe cases, respectively. Anterior and posterior lymphoscintigraphy images of each patient are routinely obtained. Therefore, we can estimate the lymphatics' positions, behind or in front, from their light and shadow in the two images instead of "guess the positions."

We divided the images obtained from 142 limbs-80 limbs in group A and 62 in group B-into five types based on abnormal patterns in lymphoscintigraphy. In addition, the relationships between the types and clinical stages as delineated by the International Society of Lymphology[6] (Table 1) were studied.

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Table 1. Clinical Stage Scale[6]

Stage 0:	Subclinical condition
Stage 1:	Early accumulation of fluid relatively high in protein content; subsides with limb elevation. Pitting may occur.
Stage 2:	Limb elevation alone rarely reduces tissue swelling and pitting. The limb may or may not pit as tissue fibrosis supervenes.
Stage 3:	Lymphostatic elephantiasis where pitting is absent, trophic skin changes such as acanthosis, fat deposits, and warty overgrowths develop.

The operations were performed under general and local anesthesia, but the latter method was excluded in this study because the number of lymphaticovenous anastomosis per limb was small compared with the former. Of the 111 patients, 31 patients with 35 limbs underwent lymphaticovenous anastomosis under general anesthesia. Of the 50 patients with types III, IV, or V, more than half are waiting for surgery and the others are undecided about surgery because of age, financial, or social problems. Several patients had lymphaticovenous anastomosis under local anesthesia and those were excluded.

To find functional lymphatics, 5% patent blue was first injected for visualization of the lymphatics in the first and third web spaces of the affected foot. In addition, indocyanine green was then injected in the same places in the latest 21 patients of this series for detection of the lymphatics through the skin as fluorescence lymphography by special equipment, Photodynamic Eye (PDE, Hamamatsu Photonics, Hamamatsu, Japan).[7] Skin incisions for the detection of the lymphatics were made at which the lymphatics could be seen through the skin by the naked eye or by PDE. Other skin incisions were made at the distal site where dermal backflow could be seen in the area where no lymphatics could be detected through the skin. We also incised the skin for detection of the lymphatic vessels at the anterior or medial side of the thigh and leg where no dermal backflow could be seen. We consider that there are some possibilities of being able to find some lymphatic vessels even in the area of no dermal backflow because lymphoscintigraphy does not always reveal every lymphatic vessel. The number of lymphatics to be anastomosed in each type was aggregated and averaged. Circumferential measurements of the affected limbs were performed at sites 10 cm above the proximal margin and beneath the distal margin of the patella and at the ankle before and about six months after surgery. Volumes of the affected limbs were approximately calculated by those measurements.

Statistical Analysis

The data were presented as mean and standard deviation. Differences in means of the clinical stage scale between the types were analyzed using a multiple comparison test for ordinal variables. Differences between types II, III, IV, and V in the average number of anastomoses were analyzed using one-way analysis for continuous variables. We used $P = 0.05$ as the critical level for statistical significance. StatMate III (ATMS Co., Ltd. Tokyo, Japan) was used for statistical analysis.

RESULTS

Types of Images in Lymphoscintigraphy

Lymphoscintigraphic abnormalities observed were the absence or decrease of inguinal lymph nodes, lymph stasis in the lymphatics, and dermal backflow. According to those observations, the lymphoscintigraphic images obtained could be divided into five types:

Type I: Recognition of obvious inguinal lymph nodes and lymphatics along the great saphenous vein, and/or lymph stasis in the collateral lymphatics. However, the number of visible inguinal lymph nodes was reduced, so they are slightly affected (see Fig. 1).

Type II: Few or no inguinal lymph nodes can be seen. Lymph stasis in the lymphatics and collateral lymphatics are observed in the thigh and leg. Small dermal backflow may be seen in the thigh (see Fig. 2).

Type III: No inguinal lymph nodes are detected. Dermal backflow can be seen in the thigh and/or leg. Lymph stasis in the lymphatics and/or collaterals is observed (see Fig. 3). The inguinal lymph nodes and lymphatics in the thigh are affected.

Type IV: Recognition of dermal backflow and lymph stasis in the lymphatics in the leg (see Fig. 4). The inguinal lymph nodes and lymphatics in the thigh and leg are affected.

Type V: No dermal backflow in the thigh and leg can be seen (see Fig. 5). The inguinal lymph nodes and lymphatics in the thigh and leg are severely affected.

In group A, five limbs were classified as type I, 15 as type II, 20 as type III, 29 as type IV, and 8 limbs as type V. In Group B, 13 limbs were classified as type I, 4 as type II, 3 as type III, 3 as type IV, and 8 as type V on the right side of the lower extremity and 14 limbs were classified as type I, 6 as type II, 2 as type III, 4 as type IV, and 5 as type V on the left side of the lower extremity. Images obtained from 3 limbs were not classified (Table 2).

As for the relationships between the types and clinical stages, most of the limbs were classified as stage 2. However, the limbs in type I tended to be classified as stage 1 and those in type V as stage 3. There were significant

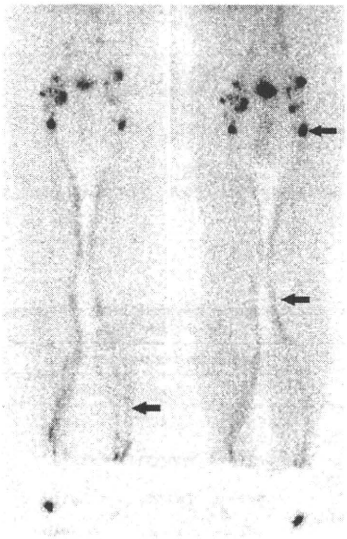

Figure 1. Images of type I in a patient with left lymphedema 30 (left) and 120 (right) minutes after injection of contrast medium. There is no obvious dermal backflow. The inguinal lymph nodes are reduced in number and lymph stasis in the lymphatics or collateral lymphatics along the saphenous vein are visible (arrows). The medium remains in the lymphatics 120 minutes later.

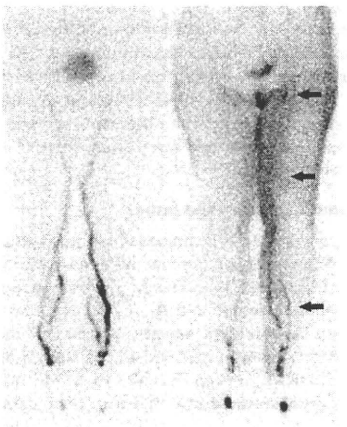

Figure 2. Images of type II in a patient with left lymphedema 30 (left) and 120 (right) minutes after injection of contrast medium. Lymph stasis in the lymphatics (arrow) and visible dermal backflow (arrow) on the left thigh can be seen. The inguinal lymph nodes are reduced in number (arrow).

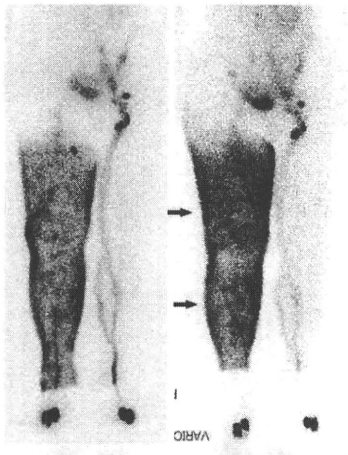

Figure 3. Images of type III in a patient with right lymphedema 30 (left) and 120 (right) minutes after injection of contrast medium. Dermal backflow (arrows) in the leg and thigh can be seen at the right.

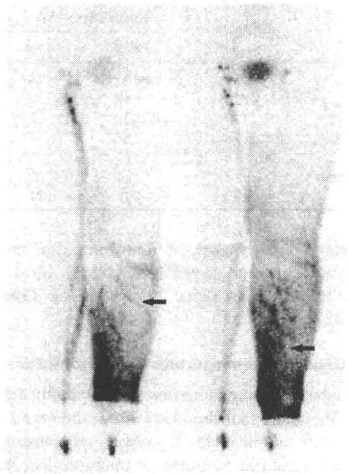

Figure 4. Images of type IV in a patient with left lymphedema 30 (left) and 120 (right) minutes after injection of contrast medium. Dermal backflow (right arrow) and lymph stasis in the lymph vessels (left arrow) in the leg can be seen and remains in the leg 120 minutes later.

Figure 5. Images of type V in a patient with left lymphedema 30 (left) and 120 (right) minutes after injection of contrast medium. There is no obvious dermal backflow in either the leg or thigh. The medium remains in the foot (arrow) and around the ankle (arrow).

Table 2. Results of Classified Types

	Group A (n = 80)	Group B (n = 31)		
		Right	Left	Total
Type I	5	13	14	32
Type II	15	4	6	25
Type III	20	3	2	25
Type IV	29	3	4	36
Type V	8	8	5	21
Unclassified	3	0	0	3
Total	80	31	31	142

differences in the means of limbs in different clinical stage scales between types I and types II to V, and between type V and types II and IV by Dunn's test (Table 3).

Detection and Anastomosis of Lymphatics

Lymphaticovenous anastomosis was applied to types II, III, IV, and V patients. However, in the type I patients and in some of the type II patients, combined physical therapy was indicated instead of microsurgical treatment because lymphoscintigraphy showed that they seemed to have some lymph pathways in the affected limb. In the 35 limbs of the 31 patients with lymphaticovenous anastomosis, the sites for skin incisions were chosen based on lymphoscintigraphy images. The lymphatics detected

Table 3. Type and Clinical Stage Scale

Clinical stage scale	1	2	3
Type I (n = 32)	19	13	0
Type II (n = 25)	3	22	0
Type III (n = 25)	0	22	3
Type IV (n = 36)	0	31	5
Type V (n = 21)	0	9	12
Total (n = 139)	22	97	20

n, number of limb; ★$P < 0.05$, ★★$P < 0.001$ (Dunn's multiple comparison test).

Table 4. Total Number and Average Number of Anastomoses Per Limb in Each Type

	Total number (average number)			
	Foot	Leg	Thigh	Total
Type II (n = 3)	3 (1)	5 (1.7)	2 (0.7)	10 (3.3)
Type III (n = 12)	13 (1.1)	30 (2.5)	10 (0.8)	53 (4.4)
Type IV (n = 16)	22 (1.4)	29 (1.8)	7 (0.4)	58 (3.6)*
Type V (n = 4)	6 (1.5)	4 (1)	2 (0.5)	12 (3)*
Total (n = 35)	44 (1.3)	68 (1.9)	21 (0.6)	133 (3.8)*

*$P < 0.05$.

were anastomosed to suitable veins in a side to end fashion.

The total and average numbers of lymphaticovenous anastomoses per limb are shown in Table 4. The average number of anastomoses per limb in type III was the highest; in the other three types, it was approximately the same. There were significant differences between type III and type IV, and between type III and type V (One-way analysis of variance, $P < 0.05$).

Volume Change of the Limbs

Excess volume of the affected limb was reduced from 83 to 3575 ml except for four limbs in which lymphedema of the affected limb became slightly worse. The average improvement of edema in all cases was 872 ± 1062 ml. The average volume reduction in each type was: 218 ± 944 ml in type II, 543 ± 616 ml in type III, 1335 ± 1288 ml in type IV, and 615 ± 817 ml in type V. There was no statistical difference among the types.

DISCUSSION

There are some relationships between the types based on the abnormal patterns of lymphoscintigrams in this study and the clinical staging. The abnormal patterns in

the lymphoscintigrams have been reported from the points of visualization of lymph vessels and lymph nodes, dilatation of lymphatic vessels, existence of collateral vessels, and dermal backflow.[1,3] These patterns vary with the progress of the lymphedema, previous treatments for edema, or the origins of the lymphedema. In cases with secondary lymphedema due to surgical intervention or irradiation therapy for oncological treatment, the lymphatic system seems to be broken from its proximal side to its distal portion. Koshima et al.[8] reported that occlusions of the lymphatic trunks and degeneration of the smooth muscle cells might start from the proximal ends of the extremities. This is quite indicative for our study and the types in this study indicate the progress of lymphedema. That is, the clinical staging increases its number as the number of the type increases.

The clinical stages only refer to the physical condition of the extremities.[6] On the other hand, the location of the edema clinically varies from thigh to foot and physical conditions do not always correlate with hypo- or dysfunction in lymph transport. However, the stages don't include these factors. A more detailed classification should be proposed according to an improved understanding of the pathogenetic mechanisms of lymphedema.

Szuba et al.[4] developed a system of lymphoscintigraphic scoring for grading the degree of lymphedema in the upper extremities after mastectomy. The score is based on visualization of the lymph nodes and extent of dermal backflow: There are 6 degrees in the visualization of the axillary lymph nodes and 4 degrees in the extent of dermal backflow. The score is correlated with the magnitude of the excess volume in the involved limb. Pecking et al.[5] analyzed lymphoscintigrams obtained from 4,328 patients and reported the staging of lymphoscintigraphic images and grading of the severity of lymphedema in the lower limbs. There are four grades of severity according to the clinical stages[6] as advocated by the International Society of Lymphology and six stages of the images classified from the aspects of lymph transport, drainage route, lymph stasis, and visualization of the lymph nodes. However, their scoring or grading systems are appropriate for evaluating the conditions of lymphedema but are insufficient for assessing microsurgical indications because they are too broad to choose specific surgical applications. Therefore, we proposed another reference in lymphoscintigraphy for microsurgical treatment.

In three cases, there were three lymphoscintigraphic images that could not be classified into any type. These images showed no lymph nodes and no dermal backflow in the leg and thigh but lymphatics in the leg, which was similar to type V, but the lymph vessels were working in the leg. Long standing physiotherapy has been applied to these cases. Conservative physiotherapy might affect lymphoscintigraphic images, particularly dermal backflow.

In our study, type III is the best indication for lymphaticovenous anastomosis because we were able to apply more than four anastomoses to one limb. There are several reasons for the facility to find and anastomose the lymphatics in type III, for example, dilatation of the lymphatic lumen, moderate thickness of the lymphatic wall, and measurable lymph flow. Although it is considered that there are many factors that influence lymphedema-preoperative conditions, postoperative nonsurgical treatments, or patients' daily activities-the number of lymphaticovenous anastomoses in one limb should be one of the factors for improving lymphedema. In this study, there was no statistical difference of volume reduction in each type. This might be attributed to the small number of cases in this series. However, Huang et al.[9] mentioned that the results after microlymphaticovenous anastomoses were proportional to the number of anastomoses. In addition, Campisi et al.[10,11] stressed that the earlier the treatment, the higher the edema reduction, and that it was important to select suitable patients for surgery. We presume areas suited for anastomosis vary according to type. The average number of anastomoses in one limb was almost the same in types II, IV, and V. However, that in the leg of type V was 1 and was the worst among them, meaning the area for anastomosis in type V might be more distal than those in types II and IV. Vaqueiro et al.[12] mentioned the relationship between the images of lymphoscintigraphy and lymphaticovenous anastomosis. Lymphaticovenous anastomoses were possible only in patients who had patent lymph channels visible on lymphoscintigrams. However, while this seems to be true, it is possible to find lymph vessels clinically even in areas of dermal backflow. Although these lymphatics are masked by the contrast medium in these areas, we could indeed find functional lymphatics and complete lymphaticovenous anastomosis in types II, III, IV, and V. Furthermore, MRI-lymphography sometimes provides good visualization of the lymphatic vessels[13] but it is difficult to obtain the best visualization because the timing required to take images varies with each patient. Development of new methods or equipment will enable the detection of masked but functional lymphatics, which may contribute to the improved treatment of lymphedema for patients with severe peripheral lymphedema.

The efficacy of the treatment for peripheral lymphedema depends on many factors, kinds of treatment, timing, and so on. Therefore, it is difficult to describe the indications clearly. A prospective study is required for a complete inquiry in the future.

CONCLUSION

From the images obtained in lymphoscintigraphy in this study, we were able to classify secondary lymphe-

dema into five types that suggest the progress of secondary lymphedema. Combined physical therapy should be indicated for type I because there are some pathways of lymph from the inguinal region to the center of the body. Both combined physical therapy and lymphaticovenous anastomosis could be applied for types II, III, IV, and V. However, for type V, it was difficult to find functional lymphatics in the leg for lymphaticovenous anastomosis. In conclusion, patients with type III secondary lymphedema are good candidates for lymphaticovenous anastomosis in the lower extremities. In types IV and V, some effort might be required to detect functional lymphatics for anastomosis compared with that in type III. Lymphoscintigraphy is a useful method for evaluating lymphedema and for obtaining indications of lymphaticovenous anastomosis in patients with secondary lymphedema.

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LYMPHATICOVENOUS SHUNT FOR THE TREATMENT OF CHYLOUS REFLUX BY SUBCUTANEOUS VEIN GRAFTS WITH VALVES BETWEEN MEGALYMPHATICS AND THE GREAT SAPHENOUS VEIN: A CASE REPORT

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Chylous reflux is a rare disorder in which chyle flows antidromically from its normal route to the extremities, thorax, abdominal cavity, or other parts of the body. We present a case of chylous reflux with megalymphatics in a 28-year-old boy who presented chylorrhea in the foot, leg, and external genitalia, lymphedema, and hemangioma in the affected limb. Lymphaticovenous shunts using subcutaneous vein grafts with valves were applied to the patient for treatment of repeated chylorrhea. After surgery, the patient has not complained of chylorrhea and been freed from conservative physiotherapy such as bandaging or application of compression stockings for lymphedema for two years. A subcutaneous vein graft with valves may be considered a useful method as a shunt between incompetent and dilated lymphatics and veins instead of a saphenous vein graft in the treatment of chylous reflux in lower extremities. We discuss these treatments based on the literature about chylous disorders. © 2010 Wiley-Liss, Inc. Microsurgery 30:553–556, 2010.

Chylous reflux is a rare disorder in which the chyle flows antidromically from its normal route from the bowel through the cisterna chyli and the thoracic duct to the extremities, thorax, abdominal cavity, and/or other parts of the body. Patients with chylous reflux are relatively young[1] and their social activities are so limited by chylorrhea, lymphedema, or cellulitis. From observations of lymphangiography, chylous reflux can be divided into two types, megalymphatics and lymphatic deficiency.[1]

We present a case of chylous reflux with megalymphatics treated by subcutaneous vein grafts with valves between megalymphatics and the great saphenous vein in a 28-year-old boy who developed chylorrhea in the foot, leg, and external genitalia as well as lymphedema and hemangioma in the affected limb.

CASE REPORT

A 28-year-man presented with congenital edema in the lower right extremity, port-wine stain on the leg and thigh, and edema of the external genitalia. Conservative physiotherapy by using bandaging and compressive garments had been applied for treatment of lymphedema for two years. Chylous discharge from the foot, leg, and external genitalia continuously repeated for the past year (Figs. 1a and 1b). The volume of the lower extremities was 4,502 ml on the right side and 3,385 on the left side. His blood test showed no abnormalities.

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Ultrasonography revealed dilated lymphatics with a diameter of about 5 mm, i.e., megalymphatics, above the ankle (see Fig. 2) and knee. Lymphoscintigraphy (Fig. 3a) and indocyanine fluorescence lymphography (see Fig. 4) also revealed dilated lymphatic vessels along the great saphenous vein and dermal backflow in the leg and external genitalia.

As the first operation to improve the chylous reflux condition in the lower right extremity, lymphaticovenous shunt operations were performed above the ankle and at the upper one third of the thigh. By harvesting a subcutaneous vein with valves from the left foot, segments of about 15 mm and 10 mm in length with valves of the vein graft were used between the megalymphatics and the saphenous vein at the thigh and at the ankle, respectively.

Each vein graft was placed with its distal end to the saphenous vein and proximal end to the megalymphatics in a fashion of end-to-side to prevent the backflow of blood (Figs. 5a and 5b). Chylorrhea disappeared soon after this operation but lymphedema of the right lower limb was not improved. Lymphoscintigraphy taken three months after the operation showed decreased dermal backflow in the lower right extremity (Fig. 3b). The patient requested additional improvement in the lymphedema of the affected limb.

Six months after the first operation, lymphaticovenous side-to-end anastomosis in the right inguinal region was then applied to reduce the edema in the external genitalia. Under general anesthesia through an incision, we could detect the subcutaneous vein and nearby lymphatics that was not similar to megalymphatics but was slightly dilated. The vein was cut and its proximal end was anastomosed to the side of the lymphatics in the manner of side to end. However, the edema of the scrotum was unchanged after surgery because the main cause of the scrotal swelling was not edema but a hydrocele that had been scheduled for treatment by urologists.

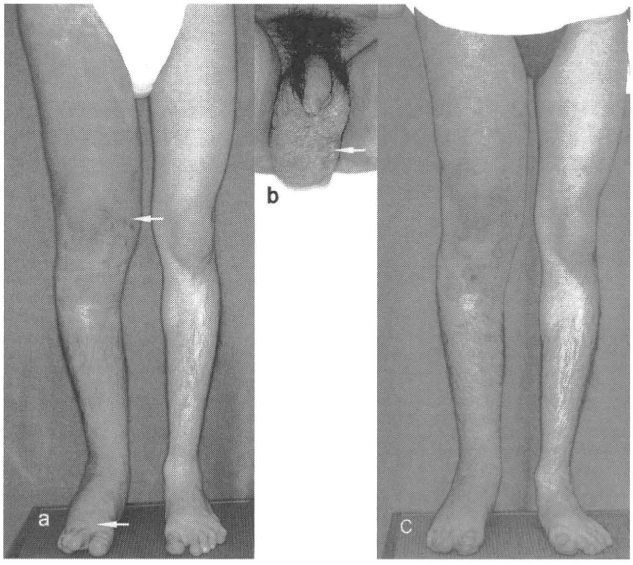

*Figure 1. (**a–c**) Frontal view of edema and port-wine stain on the right lower extremity (a) and development of a hydrocele in the external genitalia (b) before surgery. Chylorrhea was seen in the foot, knee, and external genitalia (arrows). Frontal view of the lower extremities three months after the second operation (c). No remarkable change in volume on the affected limb between before (a) and after (c) surgery. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]*

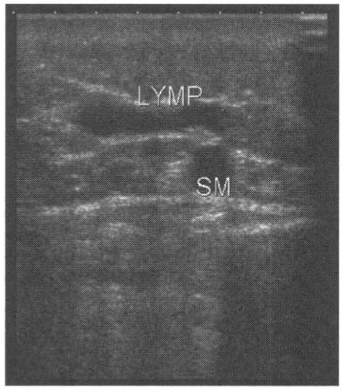

Figure 2. Ultrasonography image. Above the ankle, megalymphatics (LYMP) can be seen along the great saphenous vein (SM).

For two years after these operations, the patient has not complained of chylorrhea and has avoided conservative physiotherapy such as bandaging or application of compression stockings for lymphedema. The postoperative volume of the affected side was 4,496 ml and unchanged from the preoperative volume even though there was a weight gain of 3 kg after surgery (Fig. 1c). The patient has not developed any respiratory problems and a chest X-ray of this patient revealed no change before and after surgery.

DISCUSSION

Kimmonth et al. classified chylous reflux into two types according to observations of lymphangiography.[1] In Syndrome I, megalymphatics exist in the abdominal cavity, pelvis, or extremities with congenital naevi present on the skin and in Syndrome II, edema was often congenital and more widespread, affecting several limbs. Lymphangiography revealed few or absent subcutaneous lymph vessels. Hypoproteinemia was noted, often with other metabolic disturbances, and the long-term prognosis was poor. These kinds of lymphatic disorders have recently been diagnosed by less invasive methods such as MRI, lymphoscintigraphy,[2] or fluorescence lymphography instead of conventional lymphangiography using oily material that might cause oil emboli in the lungs.[3] This case can be classified into Syndrome I because it has the typi-