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HOW TO PLAN DECONGESTIVE THERAPY WHEN RESSOURCES ARE LIMITED. A PROPOSAL OF ALGORITHMS OF TREATMENT FOR LYMPHEDEMA PATIENTS

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INTRODUCTION

The management of lymphedema involves decongesting lymphatic pathways in order to reduce the size of the limb; encouraging the development of collateral drainage routes and stimulating the function of remaining patent routes so as to control the swelling at long-term.[1]

Traditionally, treatment in Europe has followed a "two-phase" approach: Phase I an intensive phase (decongestion phase) and a phase II or maintenance phase[2], and most of the guidelines of different countries follow this model of treatment.[3,4,5,6]

The Decongestive Lymphatic Therapy (DLT) is backed by longstanding experience and it consists of skin care, manual lymphatic drainage (MLD), range of motion exercise and multi-layered bandages[7], or as recently named "multicomponent" by the *International Compression Club*.

Its efficacy in reducing the volume in established lymphedema is well known and supported by several authors.[8,9,10,11,12,13]

There is a great deal of controversy as to which components of physical treatment programme are the most effective in reducing the edema.[14] The threshold for intervention in breast cancer-related lymphedema has been established by Specht et al. A relative volume change of 5-10% at 3 months is correlated with a progression of lymphedema volume.[15]

Current treatment of choice for lymphedema is Decongestive Lymphatic Therapy (DLT). However, DLT is not offered to patients in many centers because of the need of specialized and well trained physical therapists that can dedicate more than one hour to each patient, because of the lack of these professionals, and because of the long waiting list to be treated.

The aim of this paper is to propose a plan of treatment to deal with the limited resources that we have in Spain, and the problems of the patients with lymphedema, in order to optimize the results of the treatment.

DECONGESTIVE LYMPHATIC THERAPY

While manual lymphatic drainage (MLD) is the core of DLT, few randomised studies have been performed and they have failed to report any extra benefit with MLD.[16,17,18,19] Actually, best approach to manage lymphedema is still to be determined.[20]

The questions are, not only if MLD is useful during DLT but which kind of patient can benefit from it? Which lymphedema features are more susceptible to the MLD effect? Is DLT without MLD effective in some patients?

The recently published Cochrane review states that, MLD is safe and may offer additional benefit to compression bandaging in patients with mild-to-moderate BCRL, compared to individuals with moderate-to-severe BCRL.[21]

The use of intermittent pneumatic compression (IPC) as a component of DLT is extended but controversial. Several authors are against its use because it moves the fluid but not the proteic part of lymphedema.[22,23] Despite the report of increased genital lymphedema with IPC,[24] other studies have demonstrated its safeness and lack of adverse effects with a controlled use.[25,26,27,28,29] IPC is useful in reducing the volume of the limb,[30,31] and can be used to improve the results of DLT.[32] The use of multichambered pump has shown the best results.[33] The effect of IPC with pressures of 30-80 mmHg aid venous return, reduce edema and may even help to increase arterial flow.[34] In addition to the effects on microcirculation decreasing blood capillary filtration,[35] IPC releases anticoagulant mediators anti-inflammatory and vasoactives substances.[36] The recommendation is to monitor the possible adverse effects, employing the IPC in a hospital setting, under sanitary staff supervision.[25,26,27,28,29]

As another component of DLT, inelastic multilayer, multicomponent bandages (MB) are effective in reducing the volume of the lymphedematous limb.[37,38,39,40,41,42] This therapy has the higher degree of recommendation in the existing systematic reviews.[20,43,44,45,46,47,48] Moreover, compliance to bandages during DLT is one of the most important predictors of response, a good compliance improved the reduction in 25%.[49]

According to Kärki, although MLD is theoretically given as part of DLT, in reality, MLD is often given without compression therapy.[50]

The results of DLT reported in different papers are very variable. Excess volume reduction, calculated as a percentage of the edema reduction, varies from 22% to 73%,[1,14,21,47,51,52,53] more important in lower limb lymphedema than in upper limb.[52,54,55] Studies also report an improvement in Quality of life with DLT.[56]

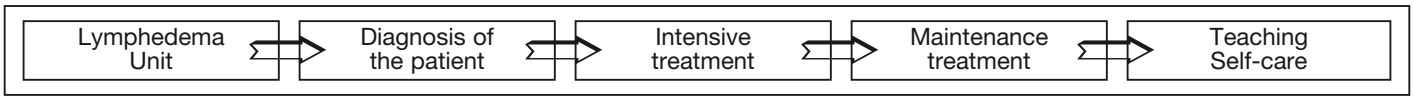
Number of sessions: Most of the edema reduction is obtained after 4-6 days of therapy.[57] In mild lymphedemas, few sessions can be very effective. A good communication between lymphotherapist and physician is necessary, in order to optimize the duration of the DLT, if a good result is obtained, maintenance phase can be started, reducing the overall duration of treatment.

HOW TO PLAN THE MANAGEMENT OF PATIENTS WITH LYMPHEDEMA

The aims of our management plan are:

1. To provide an accurate diagnosis of the disease: lymphedema, lipedema…
2. To establish a protocol of intensive treatment to reduce initially the volume of the limb.
3. To emphasize the need of self-care: teaching the patient and his care-giver to perform self-MLD, self-bandages, weight control strategies…
4. To prescribe compression garments and to check its fitting.
5. To teach to identify complications and other reasons to look for help.

Patients with lymphedema in their upper and lower limb, phleboedema or lipedema, in stages I-II (Box 3), can be treated only with education and multilayer bandages. As every treatment has to be individualized, the team has to check if MLD has to be applied, or adverse events occur. Sometimes, the patient cannot come to the hospital to receive DLT, due to the long distance from home or due to dependency from the caregiver. In these cases, we teach the bandages technique to the care-giver, in order to get a volume reduction, a more homogenous shape of the limb and to adapt more easily a compression garment. The caregiver performs the bandages at home until the reduction of volume is reached, and we check the technique and the result and we prescribe a compression garment. Good results can be reached with this treatment (Box 5). New devices with Velcro are a good option for self-bandages because they provide a better satisfaction than traditional self-bandages because of its easy doff-and-donning, and an improved

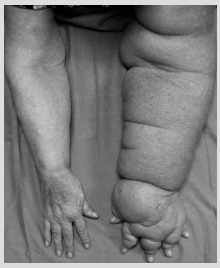


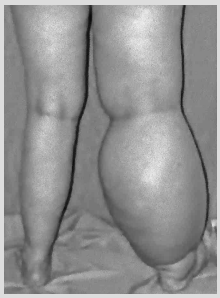
Box 1 - Management of patient in a Lymphedema Unit.

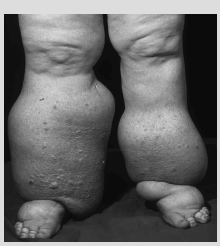
For patients with elephantiasis (Box 2), fibrosis at the root of the limb, genital or central lymphedema, facial lymphedema, children and pregnant patients, the general recommendation is MLD followed by multilayer bandages. These patients receive 10-20 sessions of MLD (45 minutes), followed by Intermitent pneumatic compression (IPC) in some cases (if it is not contraindicated), and in all of them multilayer bandages are applied and have to be worn until next session.

compliance.[58] In moderate lymphedema patients, we prescribe 10 sessions of DLT, preferably in consecutive days, at 6th or 7th day, volume is measured and if the result is satisfactory, compression garment can be ordered. The rest of the sessions can be programmed on alternate days, until the garment is ready. In severe lymphedema patients with fibrosis, we prescribe 15-20 sessions in order to obtain the greatest reduction possible.

Box 2. Management of the patient with Elephantiasis

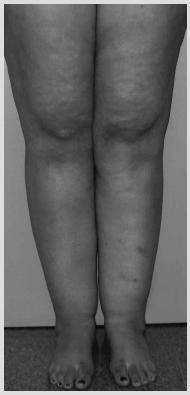






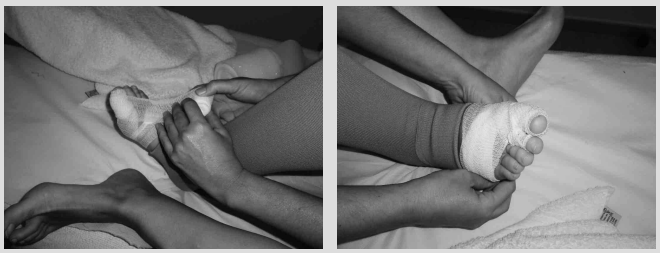
- **Inpatient management in order to avoid cardiovascular complications during treatment.**
- **Internal Medicine or cardiologist Consultation.**
- **Diuretics the first 5-7 days.**
- **Exercises in the swimming-pool.**
- **DLT: MLD, intermittent pneumatic compression if it is not contraindicated, and multilayer bandages.**
- **Skin care, exhaustive drying of the foldings, emollient cream, wounds care.**
- **Inpatient management during at least the first 7 to 10 days. The treatment has to be continued until a plateau is reached in the reduction of the volume.**
- **Flat-knitted compression garment prescription: class 2 for upper limb; class 3 for lower limb. Sometimes overlapping of different garments is a better option to control the volume and to improve the fitting.**
- **The limb must be bandaged until the garment is fitted.**
- **Teaching Self-bandages for night use.**

Box 3. Upper and lower limb lymphedema, phleboedema, lipedema, in stages I-II

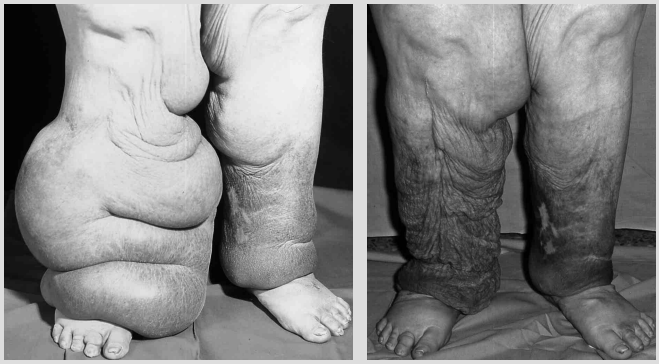


- **Self-bandage teaching.**
- **If pitting is present: 5-10 sessions of Intermittent pneumatic compression, between 30-60 mmHg with a multicompartmental device, under the team control. Followed by multilayer bandages.**
- **Exercises.**
- **Compression garment prescription: flat-knitted for lymphedema patients; Circular knitted for phleboedema and lipedema patients.**

Box 4. Self-bandage technique



Box 5. Treating patients only with multilayer bandages



- **Mild or moderated lymphedema.**
- **Without proximal involvement.**
- **Positive Pitting sign.**
- **Patients living far from the hospital.**
- **Dependency syndrome.**

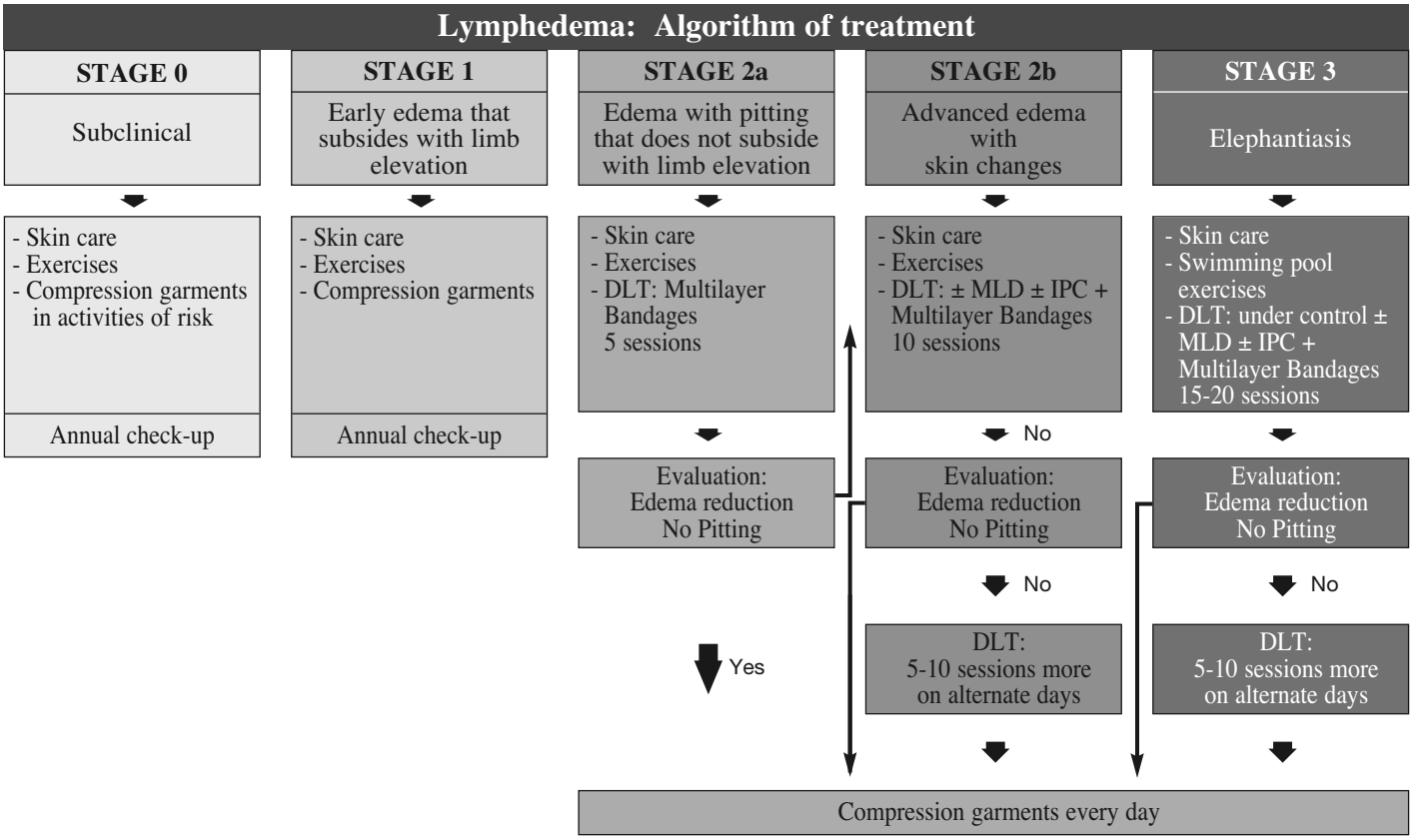


Figure 1. Algorithm of treatment for patients with lymphedema when ressources are limited.

Following Evidence Based Medicine criteria, we'd like to underline that the decision making about the therapeutic plan in a lymphedema patient has to be based on three pillars:

1. The best scientific evidence available
2. Our clinical experience
3. Characteristics, needs, values and preferences of the patient.

At the same time, the decision making is influenced by the institutional environment where we work (public hospital, private setting…).[59]

A health professional has to combine the clinical ability and experience with the best scientific evidence available, none of them is sufficient by itself. Without the experience, clinical practice could become tyrannized by external evidences, because even when these evidences are qualified as excellent, they could be not accurate or not applicable for a single patient. Without the best scientific evidence, clinical practice could become phased out, to the detriment of the patient.

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i-Press® PNEUMATIC DRAINAGE VERSUS MANUAL DRAINAGE IN UPPER LIMB SECONDARY LYMPHOEDEMA SAME COMPRESSION, SAME BENEFIT?

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Key words: lymphœdema, manual drainage, plethysmography, pressotherapy, upper limb.

ABSTRACT

Introduction: Pressotherapy is widely used but is often said to have lesser compression yield than manual drainage in upper limb secondary lymphoedema. This idea is difficult to wipe out. One of the main complaints is to find in the anterograde mode of nonprofessional material used or using. Since 1993, some pumps can work in a retrograde mode.

Objective: Our aim was to compare the effects of two light retrograde drainage options: a pneumatic and a manual one's.

Method: Retrograde pneumatic (a seven-compartment *i-Press*® *10*th serial; Electronique du Mazet, France) and manual drainage is successively and randomly carried out on 9 women (71 years old) with an old (14 years) persistent upper limb lymphoedema that appeared 7 years after radio-surgical treatment against breast cancer. All volume variations are recorded continuously with a plethysmograph (JSI, SU4). Mercury gauges are fitted 4 inches (20 cm) above the elbow. The protocol of pneumatic drainage consisted of a standardised retrograde approach with constant pressure (40 mm Hg) (without regressive pressure) at a single to double-level of compression.

Results: By use of Kruskal and Wallis, one-way ANOVA on ranks, the effect of 40 mm Hg was similar (NS) when the drainage was applied manually (0.03 ml/100 mloed/mmHg/min) or using the pneumatic pump (0.03 ml/100 mloed/mmHg/min). After 15 min stopping management, improvement mainly persisted.

Conclusion: Whatever the technique used, there is no better edema reduction at 40 mm Hg: with the help of a same retrograde mode, light drainages give the same benefit.

INTRODUCTION

Drainage of edema is the problem, not the solution! There may be a solution, a single, correct treatment for a problem that is chronic and progressive. However, the mainstay of lymphoedema management may be summarized in only three words: repetitive intermittent compression. Those goals can be obtained by physical exercises with bandages, manual drainage (MD) and pneumatic drainage (PD). That last M/PD has been reported to be beneficial in reducing upper limb lymphoedema. But little is known about their comparative effectiveness. Of course, comparing conservative management results across trials appear rapidly to be a painstaking task. It is complicated by the substantial variation of definition and stratification of lymphoedema (LO), material and regimen of DP, mode and type of additive therapies, general therapeutic strategy, and measures/mode of objectivation of outcomes among teams of researchers.

Of course in the conservative treatment, it is not possible to achieve a double blind study. On the other hand, just like drugs, an element can be compared to another. The challenge then is for the studied element to give better results than the other taken as a gold standard. To that end, the DM was taken as gold standard; the DP as candidate. Effectively, some consensual recommendations have turned MD into an indispensable technique in any decongestive method. DM seems to be duly accepted without calling in question again its effectiveness. Well, we have to leave it to a leading light in lymphology, M Földi: « it is illusory to pretend to manage a LO by single DLM » [1]. An irreversible LO gives decongestion hard time! Thus, among all goals, drainage is the most difficult to feign. In other words, clinical action speaks louder than consensus words!

But nevertheless, while the MD keeps up an easy conscience, PD has still bad press, is still controversial. Disinterest of PD is one of the most significant consequences of the thoughtless use of home pumps and misfit programs. More, the first vocation of external pneumatic compression was the DVT prophylaxis (1917 - Hartl) [2]. So PD was behind the times in lymphology and sole forward wave was available. And a backward program that starts at the swelling-front is needed in irreversible lymphoedema. Such backward pneumatic approach is used in our department since 1975 [3]. But we did have to wait 1992 to experiment a brand-new Quickels pump that may drain with forward or backward programs. Belgrado worked on the first 10 programs; Theys extended its limits to 90 programs. With such backward programs similar to the MD, it becomes possible to compare the reduction in limb size obtained by two options: a manual versus a pneumatic one's. A similar compression value remains to choose: 40 mm Hg.

METHOD

9 consecutive women (mean age: 71 y [54-83]) with monolateral (5 left; 4 right) related breast cancer arm lymphoedema (LO) were prospectively studied. The onset of LO was 21 month [7-35] after radio-surgical treatment. It is about refractory LO that runs its course without improvement or decline for the last 14 years [7-32].

The first participant was randomized to receive either MD first or PD first. Next the order of execution was permuted after each case. A 15 min rest interval is provided between each session of 16 min treatment.
The *i-Press*® device was a model 10 (Electronique du Mazet™ , France). The arm garment consists of 7 overlapping chambers. The modularly device settings (magnitude of compression, retrograde sequential mode, compression-release times cycles) were adjusted to be more closely emulating the parameters used in the manual drainage. The protocol of pneumatic drainage consisted of a standardised retrograde approach without regressive pressure (40 mm Hg) at a single to double-level of compression. The drainage initiates in the groin and then moves backwards distally with a centripetal action that ends at the groin.
To secondary compare the present results to those obtained by the use of a recent 12 chambers PD pump (Hydroven®) that generates no milking effect. The mode chosen is called "fragmentation" (Fig. 1). The 7th chamber, the highest one is inflated; the 40 mm Hg is hold during 2 s. The deflation time is about 4 s. Once deflated, the 6th chamber is inflated and deflated the same way. After 4 s, this last is once more inflated. During this holding inflation time, the 7th chamber inflates and holds pressure. Next, the 6th chamber is deflated first to the 7th one. The third wave begins and inflates the 5th. And so on, with no more than 2 chambers simultaneously fully inflated, the sequential wave begins more distally but the drainage still flows with stream. A full cycle takes 6 min 15 s.
The pneumatic pressure used was as mild as required for some MD: 40 mm Hg. To train the PT exerting a P of ± 40 mm Hg, he has to mass over a sphygmomanometric arm cuff wrapped around the non-oedematous opposite arm. The pneumatic cuff was first inflate at 10mm Hg and deflated to zero. Next, the PT placed his two hands around the cuff and presses it to reach the 40 mm Hg required. Naturally that is just an approximate value. Discrepancy with the real pressure is inevitable and partially explained with the compliance of the cuff and the fact that the manual cutaneous shear stress cannot take into account.
The *i-Press*® is equipped with two-steps intra-alveolar pressure control that is made firstly to close the valve when desired pressure is reached. This correction allows the reduction of the radius of curvature due to the reduction of the LO. The second step is made to correct the positive pressure gradient due to the inflation of the next overlapped cuff.
To assess the response of the LO, it has taken as a basis, the compliance. It expresses the relationship between the relative change in volume (% δV/V) and force (P). It should be noted that the volume value of the initial swelling (V = swollen segment volume - healthy segment volume) does not give a precise value of edema: this one is imprisoned in fibrosis and excess fat that are not mobile. These components have a very high elastance. Like a sponge emptied or filled with edema, they keep back - almost ' immediately - their initial volume. Therefore, the observation is extended by 15 minutes beyond the 16 minutes of session. During this time, there are four possibilities. The volume returns directly to the original volume; this is the case of spongiform tissues which one comes to talk. Two, tissues are more or less slowly re-infiltrated. Three, the volume remains stable and at a lower value than the initial by effective drainage of the LO. Four, the result reaches a more voluminous value by agglutination of the LO and blocking of the drainage at the root of the arm.
All volume variations were recorded continuously with a plethysmograph (JSI, SU4). Mercury gauge was fitted at the root of the arm about 4 inches (± 20-25 cm) above the elbow, at the half-width of the7th chamber. Women were seated in a comfortable chair, the upper limb lying on the table at the level of the heart (zero-phlebostatic level); the elbow extended; the fore-arm in a pronate position.
A Kruskal and Wallis, one-way ANOVA on ranks for repeated measures was used to compare the two modes of treatment.

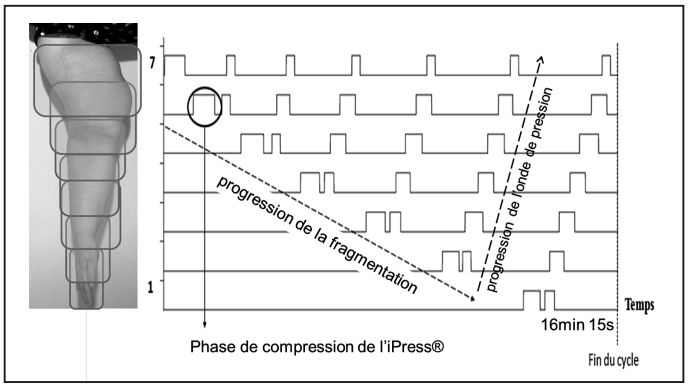

Figure 1 - Retrograde fragmentation wave progression and centripetal pressure wave progression during one cycle of *i-Press*®.

RESULTS

With a "fragmentation" mode and 40 mm Hg of compression, the volume of LO is slowly mobilised in 8/9 cases (Fig. 2). At the end of the session, there was only one case there were no volumetric variation as much by the MD as the PD. Per unit of oedema, mm Hg and time, the volumetric upper-arm decrease reaches 0.03 ml/100 mloed/mmHg/min manually and the same by means of PD. No case did show an upper-arm volumetric augmentation that reveals a local blockade and engorgement of LO. After 15 min stopping sessions, improvement mainly persisted. At the end of the sessions (16 min), the volumetric reduction does not soften. Those preliminary results do not isolate any influence of the succession of the MD/PD sessions (ANOVA; p = 0.571).

DISCUSSION

The 40 mm Hg retrograde wave yield was similar when the D was applied manually or using the pneumatic pump. These data do not support the hypothesis that PD produces lesser efficacy than MD. On the contrary, our results confirm the observations of Forner [4] and entitle to believe that the pumps or program studied in other studies evidently were not adapted to old upper-limb LO like studied here.
Conversely, our results need to be viewed cautiously. They may start the diffusion of rumours like: how is it possible to continue DP while a MD may results at the same level of results? Such distracted lector did forget that one of the main secrets of decongestion is the length of the compression/day: Edema

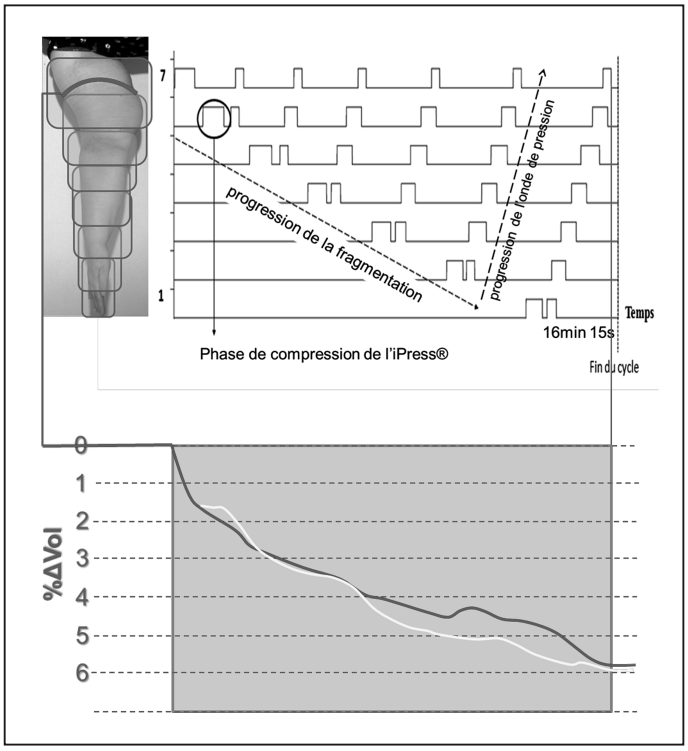

Figure 2 - Progressive mean relative arm volume reduction (%ΔVol; dark line) during a cycle of pneumatic fragmentation (*i-Press*®) at 40 mm Hg. In comparison with the result obtained by manual drainage (retrograde approach, centripetal pressure wave of ≤ 40 mm Hg).

reduction rate must be greater than the daily accumulation of edema. In recurrent LO, 1-2 h/day drainage are insufficient compared to the remaining 22-23 h without. At most, 1-2 h/session/day is just limited to cover the daily accumulation of edema. With such poor efficiency, it is not surprising Földi concluded that 'solely, any 30-40 mm Hg MD will never reduce a LO' [1]. Greater decongestion would be finding with higher pressure [5-7] or longer sessions [6] that can be obtained by cheaper PD.

But the fact that the value of 40 mm Hg has show an action in our old LO does not make it the optimal pressure, the ideal one. This one is equal to *33 torr* [8-9]. The talent of this equation formulated by Vodder is to make believe at a quantitative value while it has none [10]. Indeed, in the mouth of a Vodder who was interested in the study of the languages, the number 33 could be without value. In fact, Vodder said 33 as the doctor asked the patient "say 33" to examine the back of his throat. In fact, the patient has not to say the number 33 but 'no matter what'; therefore, he responds «aaaaah».

Thereabout for Vodder, "33" is not a numerical reality just as 'ideal' is in no way a restrictive connotation [10]. Similarly, *torr* should not be understood in the letter. Already in his time, the Torricelli had no right of citizenship in recognized pressure units [10]. On the other hand, *torr* symbolized the Norse God, Thor with its magical hammer; a hammer that is used to exert pressure... Besides after the statement of his equation, Vodder explicit this non-quantitative notion of the ideal pressure: "the ideal pressure should be great enough to produce the greatest therapeutic result." [9]

In other words, the optimal pressure measurement, it is up to what force it wins in drainage. And this measure remains unformulated and undeterminable. Indeed, the drainage requires continual interaction of four major classes of variables: the force, the duration of compression, the consistency of edema and the resistance of the surroundings tissues (thickness and fibrosis) [6,11].

But this is in accord with a second degree reading of the Vodder formula. Today, this double meaning has lost its symbolic meaning and the tracing of the number was derived from its original meaning: *"33 torr"* was changed in *30-40 mm Hg*. Which "law" you choose do not depend somewhat on the meaning you want to convey. The restrictive margin of pressure (30-40 mm Hg) has only one function: to secure the prescriber [10] because the DLM and the PD are practiced by anyone, who doesn't have sufficient knowledge of the pathology to be treated [4]. This is the reason for our choice set to 40 mm Hg.

CONCLUSION

1. In late-stage old refractory arm lymphedema, a light (± 40 mm Hg) intermittent compression do result in a small yield of 0.03 ml/100 mloed/mmHg/min.
2. The manual and the *i-Press*® pneumatic fragmentation both provide the same reduction of arm volume.
3. No case did show any engorgement of LO at the root of the arm.
4. After stopping sessions, improvement mainly persisted.

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DIAGNOSTIC, THERAPEUTIC AND ASSISTENTIAL PROJECT (PDTA) FOR PRIMARY LYMPHEDEMA

Piedmont and Aosta Valley Regions (Consortium for Primary Lymphedema of Piedmont and Aosta Valley)

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ABSTRACT

Primary lymphedema is a rare and chronic disease caused by an inherent defect in the lymphatic vessels or lymph nodes.
The final diagnosis is clinical, but in many cases it is very difficult to differentiate primary lymphedema from secondary lymphedema or from other causes of chronic limbs swelling, such as venous insufficiency.
This article focuses and describes a standardized operational approach developed from 2009 and currently in use in two Northwest regions of Italy, respectively Piedmont and Aosta Valley.

INTRODUCTION

Primary lymphedema is a chronic, evolutive and debilitating pathology of the lymphatic system, linked to a congenital lymphangioadenodysplasia / aplasia of the lymphatic vessels and/or of the lymph nodes, which is manifested by an abnormal and progressive accumulation of lymph in interstitial tissue, initially localized and subsequently extended [1]; while it is most prevalent in the lower limbs, the upper limbs, the genitals or the face may also be impacted.
The prevalence, according to the data of the international literature [2], is estimated at 1 case out of 6000/10000 individuals, with a M:F ratio = 1:3.
A distinction between hereditary and idiopathic forms can be made, according to Földi [3], in relation to the time of onset of edema (Table 1).

Hereditary Lymphedema	Early (S. Nonne - Milroy: present at birth)
	Late (S. Meige)
Idiopathic Lymphedema	Congenital: present at birth
	Early: outset before 35 years of age
	Late: outset after 35 years of age

Table 1. Classification of primary lymphedema according to Földi.

The classification proposed by Földi and ISL [1], the International Society of Lymphology, was adopted for the staging of the disease (Table 2).

Stage 0	Latent lymphedema (detectable only by lymphoscintigrafic examination)
Stage 1	Spontaneously reversible, smooth texture, positive fovea sign
Stage 2	Spontaneously irreversible, hard texture, positive fovea sign
Stage 3	Elephantiasis, severe limb deformity, fibrosis and fingers papillomatosis

Table 2. Lymphedema Staging according to Földi - ISL.

In 2009, within the activities of the Regional Coordination Centre for Rare Diseases (CMID), the multidisciplinary and multiprofessional Consortium for Primary Lymphedema of Piedmont and Aosta Valley regions was created, composed by angiologists, vascular surgeons, physiatrists, physiotherapists, nuclear physicians, infectious disease specialists, all operating

within the Public Facilities of the Italian National Healthcare System.
A Diagnostic, Therapeutic and Assistential Project (PDTA) was created by the Project Team, with the following purpose:

– Define clinical, instrumental, homogeneous and shared diagnostic criteria;
– Define therapeutic-assistential paths, appropriate to all stages of the disease;
– Create an assistential network, coordinated at regional level;
– Collect data for epidemiological and research purposes.

THE PDTA: DIAGNOSTIC CRITERIA AND THERAPEUTIC CRITERIA

DIAGNOSTIC CRITERIA

Clinical elements

The Primary lymphedema diagnosis is essentially clinical, but it often requires an instrumental confirmation. Therefore it is essential to proceed with a thorough medical history to exclude lymphedema generated by surgery, radiation therapy, presence of neoplasia with secondary involvement of local-regional lymph nodes, trauma, venous edemas or lipoedemas.
The objective examination includes inspection, palpation and search for the Stemmer sign, the indirect edema volumetric measurement [4] according to the formula of the truncated cone "frustum method" limb V = S p (X2 + Y2 + XY) h / 3 (where X is the circumference at the most distal point of the measured limb segment and Y is a point set at 'h' cm above point X).
Unilateral lymphedema can be rated "mild" (volumetric difference < 20%), "medium" (volumetric difference 20-40%) or "severe" (volumetric difference > 40%).
The patient is initially evaluated by an angiologist or a physiatrist, who, in case of suspected primary lymphedema will report the patient to the Regional Registry of Rare Diseases, for temporary exemption from health care costs for definitive diagnosis checks.

Laboratory data

The following parameters are normal in primary lymphedema: creatininemia, urine test, 24-hour proteinuria, hepatic enzymes, TSH, pro-BNP or NTpro-BNP, Reumah test, VES, PCR.

Instrumental elements

According to the Italian Society of Vascular Diagnostics – SIDV protocol [5], the instrumental framework of the patient can not leave aside a venous EcoColorDoppler ultrasound evaluation of the limbs with lymphedema (and if necessary dynamic EcoColorDoppler), in order to exclude venous thrombosis, thoracic outlet syndrome, and arterio-venous malformations.

Complementary investigations

Complementary investigations are to be considered in cases of doubt: upper and lower abdomen echography, chest X-ray, ECG and, if necessary, TAC/RMN.

Although Lymphoscintigraphy is generally accepted as the elected investigation for diagnosis and confirmation of primary lymphedema, its main limitation is the absence of procedure standardization. With the exception of a few Centers in Italy that can boast a long and consolidated experience in the scintigraphic lymphedema diagnosis, there is generally a lack of interest by the Nuclear Physicians into a rare disease such as primary lymphedema. The main consequence is an inadequate / insufficient knowledge by the majority of Nuclear Physicians on both anatomical and physiological characteristics of the lymph transport system and of qualitative / semi-quantitative scintigraphic patterns. It is on this basis that the three / five stages Lymphoscintigraphy according to the protocol of Pierre Bourgeois [6] was identified by the Project Team as the mandatory procedure for Nuclear Medical Services under the Consortium. This protocol, undoubtedly more challenging compared to others, has become mandatory in Belgium, it is extensively validated in the literature and has, above all, an educational value for Nuclear Physicians.
In the case of previous scintigraphic studies performed with other protocols or diagnostic doubts related to the test quality in pediatric patients, patients will be subject to nuclear medicine specialistic evaluation, to decide on the reliability of such scintigraphic assessments and on the need to repeat the analysis according to the Pierre Bourgeois protocol or, whether a scintigraphic examination check is required in case of a significant clinical picture variation overtime.
At the end of the diagnostic path, provided that all eligibility criteria are duly met, prescription and temporary exemption report will be converted into definitive report.

THERAPEUTIC CRITERIA

Pharmacological Therapy

At this point there is no evidence on the possibility to modify the course of the disease using oral benzopyrenes, gamma-benzopyrones, etc.. Diuretics are not advised in the treatment of primary lymphedema [2,3,7,8].
Conversely, the treatment of bacterial or fungal infectious complications is fundamental, with antibiotics (amoxicillin per os, intravenous ampicillin, fluoroquinolones, second-generation cephalosporins, tetracycline) or antifungals (fluconazole or terbinafine), for a variable period, depending on the clinical severity, from two to four weeks. After the second infectious recurrence, prophylaxis with benzylpenicillin 1,200,000 U im every four weeks is advised (if useful and well tolerated, can last for years).

Surgical Treatment

Surgery is controversial and advised in limited cases [7]. After the operation, however, rehabilitation is still needed.

Adjuvant Therapies

Control of body weight improves lymphedema evolution, although the effectiveness of specific dietary regimens is not currently demonstrated.
In specific cases psychological support may be appropriate.

Rehabilitation Treatment

The physiatrist is the guarantor of the patient's rehabilitation prescription and path, through the development of an Individual Rehabilitation Project (PRI), which provides: clinical and functional assessment, disability degree assessment (also using specific rating scales such as, for example, ICF Classification [7]), identification of clinical priorities and rehabilitation objectives (short, medium and long term), prescription of specific rehabilitation programs in the appropriate therapeutic setting, drug therapy prescription, orthoses and assistive devices (bandages and elastic-compressive braces, footwear and orthotics, walking aid tools etc.). Elastic-compression devices may also be prescribed by a specialist angiologist or vascular surgeon.
Rehabilitation treatment adheres to an integrated therapeutical approach, supported by physiatric and physiotherapic counseling, divided into two phases :

A. *First phase* - Functional Recovery, based on the Complex Decongestive Therapy, namely: skin care, manual lymphdrainage according to different methods, multi-layer bandaging with specific materials, decongesting exercises [2,3,4,7,8,9,10,11,12]. Treatment sessions frequency can vary from 3 to 5 days / week, with constant bandage; treatment cycles may be repeated according to the patient's clinical needs.

B. *Second phase* - Obtained Results Maintenance: the patient acquires selfcare autonomy, hygiene standards of living with preventive and therapeutic objective, training on self-draining, self-mobilization, self-bandaging and on day-to-day use of adequate elastic-compressive device, specific physical exercise programs potentially included in suitable extra-medical paths of Adapted Physical Activity (AFA).

FOLLOW UP

In stabilized cases, performance of clinical checks at least once a year is recommended; the number and frequency of monitoring visits can vary in the sub-acute phase or in presence of complications. In special cases, biohumoral or instrumental polyspecialistic investigations can be performed.

DISCUSSION

According to the Rare Diseases Regional Registry, in the years 2012-2013 prevalence of primary lymphedema in Piedmont was less than 1/100,000, confirming the classification of rare disease nosology. In 2012 the number of patient reports created – temporary and/or permanent – were 23, 42 in 2013, with 37 permanent reports.
The Regional Health System currently provides to patients temporary or permanent exemption all diagnostic services, rehabilitation treatment, assistive devices and orthotics included in the National Official Card, although unfortunately with the exception of bandages and elastic-compressive braces. This latter aspect is particularly problematic since the elastic restraint is the cornerstone of the therapeutic principles of lymphedema. Hopefully in the future these devices will also be reimbursed by the Regional National Health System.
Lastly, it is important to highlight that exemption from primary lymphedema health care cost is currently only granted to patients who reside in Piedmont or Aosta Valley regions, while the rest of the Italian country is not covered by the program.

CONCLUSIONS

The PDTA for primary lymphedema is a useful tool to standardize healthcare services delivery criteria at Regional level.
The strictness of diagnostic classification serves two objectives: (1) excluding secondary lymphedema causes, otherwise unrecognized and (2) optimization of human resources, equipment and budget management.
The standardization of diagnostic methods and of therapeutic modalities will allow for support efficacy studies and application of Evidence Based Medicine criteria in this field, till very nebulous at present.
It is our hope that the regional patients reporting system will contribute to the correctness and update of epidemiological data and that our project will be extended to the rest of Italy.

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LYMPHOLOGY DEVELOPMENT IN ANDALUCIA

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Keywords: lymphedema, lymphedema unit, Andalusia

INTRODUCTION

Lymphedema is the less common form of edema, which is defined as the collection of interstitial fluid rich in proteins and inflammatory cells due to an abnormality in the flow of the lymphatic system. It is usually an irreversible process with no curative treatment available. It is classified into primary (or congenital) and secondary (or acquired) lymphedema. In the industrialized countries, the most common cause of acquired lymphedema is axillary lymphadenectomy (due to systemic disease and /or radiotherapy in breast cancer), causing the development of edema on the upper limb [1,2,5]. It is recommended to establish Lymphedema Rehabilitation Units because Lymphedema high prevalence and impact on quality of life of patients and thus provide appropriate patient care that includes: early detection, primary and secondary prevention and treatment with effective protocols (agreed by all professionals involved in this pathology). Within these units, the prevention comes from what its called "School of Lymphedema", which aim is to instruct affected or at risk patients [3,4]:

- Information and training of anatomy, physiology and pathophysiology of the lymphatic system
- Hygienic-dietary measures aimed to prevention: posture, hygiene and personal care, protection of the upper limb to any external aggression (pressure, heat, abrasions, punctures, etc…)
- Teach active specific kinesitherapy: by active exercises of the affected limb (with or without acupressure garment) and breathing exercises.
- Early identification of lymphedema and its complications.
- Self-care measures once the lymphedema: drainage, dressing,…

According to several studies, all information and educational interventions for patients, provided by the staff of the Lymphedema Rehabilitation Units, can be made at three levels [4]: *preoperative* o before radiotherapy; *immediate postoperative,* providing information to patients about postoperative pain, movement limitation of the shoulder joint, scar retraction, rehabilitation exercises, early recognition of lymphedema symptoms; the final level is to *set realistic expectations* regarding its process, treatment and evolution with the aim of reduce the feeling of powerlessness and/or frustration.
In Andalusia began the first lymphedema rehabilitation units about 15-20 years ago (we have not been able to establish the exact dates on some of them). The progressive development in recent years, the available techniques and the action protocol differ depending on the level of the Hospital (I, II, III level) where the Units are implanted.
To collect as much information as possible, the following study was carried out.

OBJECTIVES

The aim of our study is to describe the current situation of Lymphedema Rehabilitation Units in Andalucia, that is, the number of existing units, its organization and functioning.

MATERIALS AND METHODS

A cross-sectional descriptive study was carried out.
The data were obtained between June 2012 and February 2013, through a survey specifically made for this study. (Annex I)
Depending on the availability and working schedule of the surveyed, the survey was self-administered, answered by email or by phone.
The Rehab Doctor, in charge of the Lymphedema Unit, was the person who fills the survey in most cases. Only in some cases, when we couldn't contact with the Rehab Doctor in charge, the Chief Physician of Rehabilitation Service answered the survey.
46 public healthcare centers, of the 47 existing centers of Andalusian Community, were surveyed. It was decided not to ask privates healthcares centers due to the risk of bias (by collecting data of the existing center via the internet) and the reliability of the data provided.
The variables were chosen with the objective of describe the organization and functioning of the Lymphedema Units.
The Statistical Analysis was made by the statistic package of SPSS vs.20. Frequencies and average were obtained.

RESULTS

46 public healthcare centers of Andalusian Community were surveyed. We obtained responses from 43 of them. The causes of no response were the following: in one case the person in charge was off work during the study, in other case the healthcare center was recently opened and the Rehabilitation Service still doesn't work properly, and in the last case the healthcare center could not be contacted.
We obtained responses from 43 centers, and only 11 centers have Lymphedema Rehabilitation Unit, what entail that 74.41% of centers surveyed don't have Lymphedema Unit.

All units are formed by a rehabilitation team, without involvement of other specialties. 45.5% composed of a rehabilitation physician and a physiotherapist and only in one case the rehabilitation team was composed by a rehabilitation physician, a physiotherapist and a nurse.
Coordination is carried out by the rehabilitation physician in all units with part-time work in all cases. The physiotherapist, in 54.5% of the units is working full-time.
Concerning the types of lymphedema attended, 63.6% of the units cover all types of lymphedema and one unit cover only secondary lymphedema.
The medical consultation takes place an average of 4 days a month, and the follow-up of the patients is every six months in 82,8% of the units. An average of 15 patients for the first time and 32 for review are seen per month.
Regard to the treatment, an average of 20 session are applied within a specific room (90,9%) and the treatment is repeated annually in 72,7% of the units. Furthermore, 100% of unit's treatment is based on complex physical therapy (manual lymphatic drainage, compression bandaging, decongestive exercises, skin care).
And finally, only 8 of the 11 Rehabilitations Services surveyed with Lymphedema Rehabilitation Unit have "School of Lymphedema". The most common alternative for the "School of Lymphedema" is general recommendations (preventive measures, self-care measures, early identification of lymphedema symptoms and signs, and lymphedema complications) given in medical consultation or before the patient leaves the hospital (in 26% of the cases).

CONCLUSIONS

The existence of Lymphedema Rehabilitation Units allows carrying out effectives and efficent programs of prevention and early treatment for this chronic pathology. To carry out these programs successfully, high specialization of all professionals who constituted the unit is required.
In our daily clinical practice we can observe that the diagnosis of Lymphedema is made earlier when patients are included in programs such as "School of Lymphedema". This early diagnosis provides the chance to improve or/and to have a better control of the disease.
The creation of "Schools of Lymphedema" allows patients to learn more about Lymphedema by knowing the triggers and aggravating factors of this pathology. In this way the risk of the undesirables practices could be limited [4].
For these reasons is recommended to establish in all Rehabilitation Units of our Community the "School of Lymphedema" as an example of good clinical practice.
If thus is necessary to continue implementing Lymphedema Rehabilitation Units in all services of Physical Medicine and Rehabilitation Andalusia without them.
It also important to continue developing International treatment protocols of Lymphedema for the Units that are currently working and those which are planned to work in the future.

ANNEX I

LYMPHEDEMA QUESTIONNAIRE

Hospital: _____

Person in charge with who contact: _____

Contact telephone number: _____

1. Has your Rehabilitation Service a Lymphedema Unit? **(if your answer is "no" go to question number 16, if your answer is "yes" continue and leave question number 16 and 24 blank)**
2. Is the Lymphedema Unit made up of only by a rehabilitation team or is there the involvement of other specialties?
3. Who are the professionals who are part of the Lymphedema Unit? (number of person and professional category, e.g. nurse, rehabilitation doctor, physiotherapist…
4. Who coordinated the Lymphedema Unit?
5. Are the professional involved working full-time o part-time?
6. What type of lymphedema is seen? (e.g. primary lymphedema, secondary lymphedema, lipedema, …)
7. Medical consultation program (days per week or month…)
8. How many patients are seen for first time per month?
9. How many patients are seen for review per month?
10. How often patients are reviewed?
11. Where the treatment of lymphedema is applied? (specific room or shared room)
12. How many sessions of therapy are applied?
13. How often the treatment is applied? (e.g. 1 or 2 time per year)
14. What type of treatment is made?
    a. Complex physical therapy (CPT)
    b. Only manual lymphatic drainage (MLD)

➔

c. Only lympho drainage pump
d. Compression bandaging
e. Others: decongestive exercises, skin care

15. Is it prescribed a custom-made pressotherapy garment? How often they are renewed?

16. **If your Rehabilitation Service doesn't have a Lymphedema Unit**, do you know if there is any type of lymphedema treatment in your Rehabilitation Service? **(If your answer is "no", go to question number 24)**

17. How is this organised? (e.g. patient are seen in the same medical office, they are seen for the same rehabilitation doctor, the same day,…)

18. What type of lymphedema is seen? (e.g. primary lymphedema, secondary lymphedema, lipedema, …)

19. Average of patients seen per month?

20. Where the treatment of lymphedema is applied? (specific room or shared room)

21. How many sessions of therapy are applied? And how often the treatment is applied?

22. What type of treatment is made?
    a. Complex physical therapy (CPT)
    b. Only manual lymphatic drainage (MLD)
    c. Only lympho drainage pump
    d. Compression bandaging
    e. Others: decongestive exercises, skin care

23. Are you planning to set up a Lymphedema Unit shortly?

24. Finally, is there a "School of Lymphedema" in your healthcare center? If your answer is "**no**" are your Rehabilitation Service carried out any prevention measure after breast cancer surgery?

25. If your answer is "**yes**" how does this "School of Lymphedema" work? Who is the person in charge? And how often they work?

COMMENTS/CONTRIBUTIONS:

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TWO-YEAR FOLLOW-UP OF TEMPORAL CHANGES OF BREAST EDEMA AFTER BREAST CANCER TREATMENT WITH SURGERY AND RADIATION EVALUATED BY TISSUE DIELECTRIC CONSTANT (TDC)

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ABSTRACT

Background: Breast edema is reported as a common complaint after breast conserving surgery and radiotherapy (RT). Measurements of local water in skin and upper subcutis with tissue dielectric constant (TDC) technique have the potential to detect breast edema in patients after breast cancer treatment.

Objective: The purpose of the present study was to examine development of edema in breast, axilla and upper arm in women treated with breast conserving surgery and RT during a 2-year follow-up.

Method: Sixty-five patients have been included and measured at 10 time-points (before RT, three time-points during RT, 2 and 4 weeks after RT and then 3, 6, 12 and 24 months after RT). Breast edema was measured by tissue water content in skin and upper subcutis at both sides with MoistureMeterD. TDC, directly proportional to tissue water content to the effective depth of 2.5 mm, was evaluated. Definition of breast edema was determined as a TDC ratio ≥ 1.40 between the treated and healthy breast.

Results: The TDC measurements demonstrated breast edema already before RT. The mean TDC ratios at three weeks of RT and at 3 and 6 months post-RT were exceeding the edema threshold limit (i.e. TDC ratio ≥1.40). The largest proportions of patients exceeding the edema threshold limit were found at three and six month post-RT (63%) and the smallest proportions at two years post-RT (28%). Concerning axillary dissection or sentinel node biopsy, no statistically significant differences were found between the groups at any of the 10 different measurement time-points.

Conclusion: Cancer treatment related edema in the breast is very frequent at three to six months after RT but decreases at one to two years after RT. Differences in the surgical procedure are unlikely to change the incidence of breast edema during a two-year follow-up period.

Keywords: Breast edema, breast surgery, radiotherapy, tissue dielectric constant

INTRODUCTION

Breast edema is reported as a common complaint after breast conserving surgery and radiotherapy (RT) with a prevalence varying between 0 and 90% [(1)]. Breast edema is usually assessed by using clinical signs such as erythema, orange peel syndrome, and the patient's complaints of a feeling of swelling, tension, heaviness and pain [(2)] which are often found in patients with manifest breast edema. However, post-treatment changes may be diffuse depending on the extent of surgery and radiation treatment fields. Besides, these symptoms are not eventually observed in the mild breast edema [(3)].

Although mammography is primarily used for the detection of breast cancer, breast edema may be depicted on mammogram due to increased density of the breast tissue and skin thickening [(4)]. However, this method is associated with an additional radiation exposure to a very sensitive breast tissue and a non-radiation alternative is of a great value. Ultrasound was used before for this purpose and the diagnosis of breast edema was based on

measurement of dilated lymphatic channels underneath the skin and fatty parenchyma [5]. Even if this modality is significantly better than clinical assessment of breast edema at the end of RT [6], it is not the case before and during RT.
Another technique that could provide useful information for characterization of breast edema in breast cancer treated patients is TDC. The major advantages of this method are lack of radiation exposure, easy and repeated quantitative measurements of local tissue water content [7].
The aim of the present study was to investigate the temporal pattern of tissue water content in skin and upper subcutis until 2 years post-RT in women treated for breast cancer with breast conserving surgery and RT to the breast.

MATERIALS AND METHODS

Patients

Sixty-five breast cancer patients treated with breast conserving surgery and sentinel lymph node biopsy (SNLB) or axillary lymph node dissection (ALND) and RT to the breast at the Department of Oncology at Skåne University Hospital, Sweden, were included in this study. Previously, Johansson et al. (2014) recruited 118 patients but finally 65 patients to whom all measurements were performed, were included in this study. The following inclusion criteria were utilized for patient selection: female, over 18 years of age, unilateral breast cancer, undergoing breast conserving surgery, RT and SNLB or ALND. The exclusion criteria were patients with preoperative chemotherapy, breast tumor recurrence, diseases that may complicate the measurement of edema such as dementia.

Measurements

TDC (Tissue Dielectric Constant): The TDC values were measured on aff ected and contralateral sides using the MoistureMeterD (Delfin Technologies Ltd, Finland). The device transmits a very high frequency electromagnetic (EM) wave of 300 MHz into an open-ended coaxial probe in contact with the skin. A major part of the EM energy is absorbed by tissue water while the rest is reflected back to the coaxial line and an electrical parameter, tissue dielectric constant (TDC), directly proportional to tissue water content in skin and upper subcutis, is calculated [8]. With this technique local tissue water was measured to the effective depth of 2.5 mm.
TDC measurements were performed at both sites in the breast and the upper arm with the patient in a supine position, and in the axilla with the patient in a frontal position, arms along the body. Three repeated measurements were made at each measurement point and averaged.

Breast: Each quadrant of the breast was measured with the probe placed in the middle of each quadrant with the edge of the probe 10 mm from the areola (Fig. 1). The quadrant measurements for each breast were average (with exclusion of quadrant(s) with scar tissue) (2).

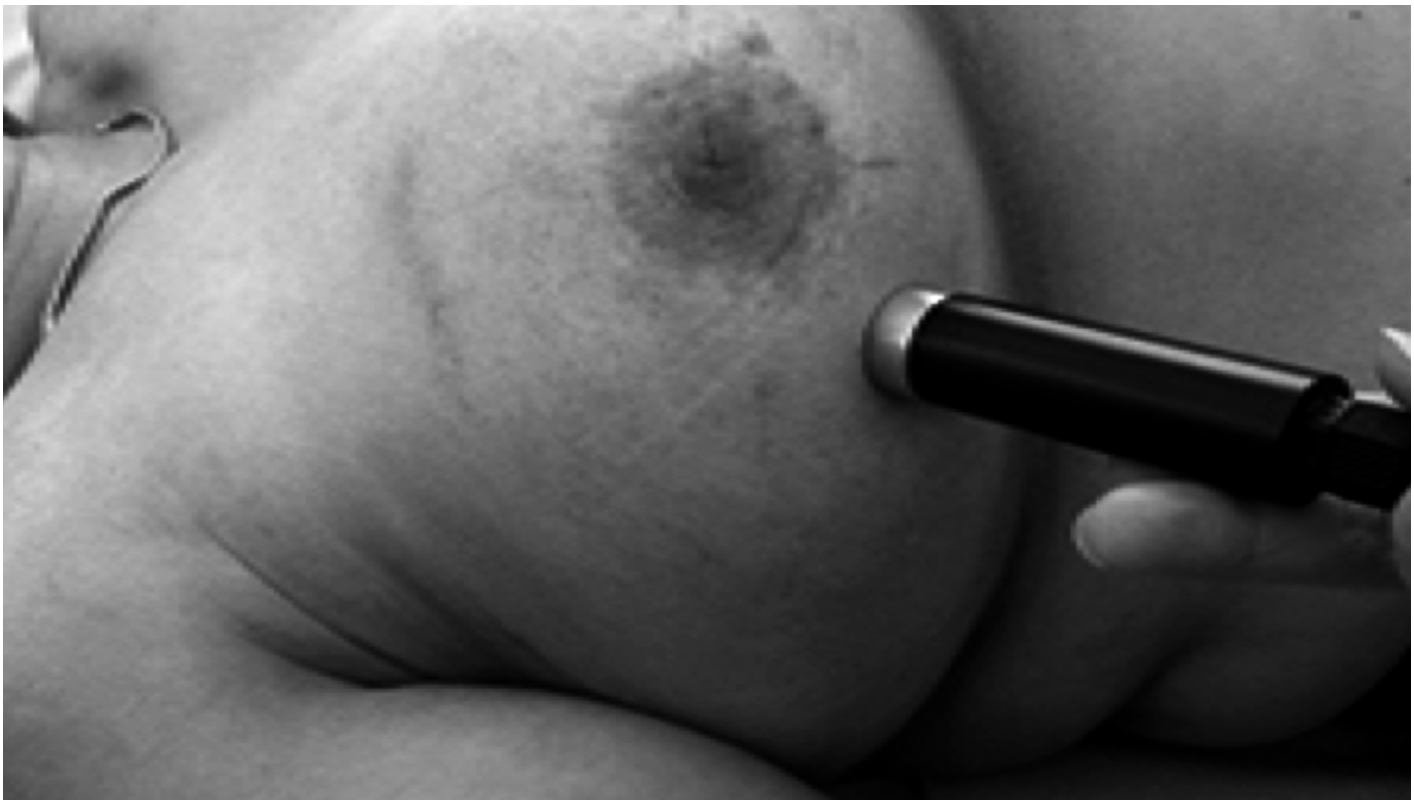

Figure 1 - Each quadrant of the breast was measured with the probe placed in the middle of each quadrant with the edge of the probe 10 mm from the areola.

Axilla: The axillary measurement site was defined at a spot 5 cm below (caudal) from a line drawn between the highest point of the fold between arm and body, and the lateral scapula edge (Fig. 2). The arm measurement was performed in the medial upper arm, 5 cm proximal to the anticubital fossa.

To eliminate individual differences in tissue water content the TDC ratio between the affected and healthy side for each patient was calculated. The TDC threshold ratio for breast edema was defined from the previous study [2] as a TDC ratio equal to or greater than 1.40.

BMI (Body Mass Index): At the baseline, the height of the patients was measured and body weight was measured at each measurement occasion to calculate changes of BMI during the follow-up period. BMI was expressed in kg/m^2.

Design and procedure

TDC measurements were made within a week before start of RT, at the end of each week of RT, at two and four weeks after RT, and at three, six, twelve and twenty-four months after RT.

Statistical analysis

TDC values showed a normal distributed and therefore independent t-test was used for comparison between TDC values in the operated and healthy breast/axilla/arm at each time-point. Since TDC ratios at each time-point showed a significantly non-normal distribution ($p < 0.01$), tests for significance of overall TDC ratio changes were done using nonparametric tests: Friedman ANOVA test was used to reject or accept the null hypothesis (i.e. there are no differences between the breast TDC ratios in all time points). Afterwards Wilcoxon matched pairs test was used in order to find in which time-point the breast TDC ratio was significantly higher than other time-points. Man Whitney U-test was used in subgroup analysis i.e. how the distribution of the TDC ratios in the ALND and SLNB groups looks. Fisher's exact test was used to find eventual differences between proportions in these subgroups. Repeated measures ANOVA was used to test the equality of BMI means. All statistical tests were done using SPSS version 22. A value $p < 0.05$ was chosen as significance level.

RESULTS

Patients

Characteristic features of the patients are summarizes in Table 1. The majority (84.6%) of the patients had lumpectomy with SLNB and the rest (15.4%) had lumpectomy with ALND. The majority (84.6%) also received hypo-fractionated radiotherapy to breast up to 42.5 Gy and 13.8% received conventional fractionation up to 50.0 Gy, and one patient (1.6%) was given a dose up to 66.0 Gy. Before RT, 73.3% of the patients were overweight or obese with no significant change during the follow-up period.

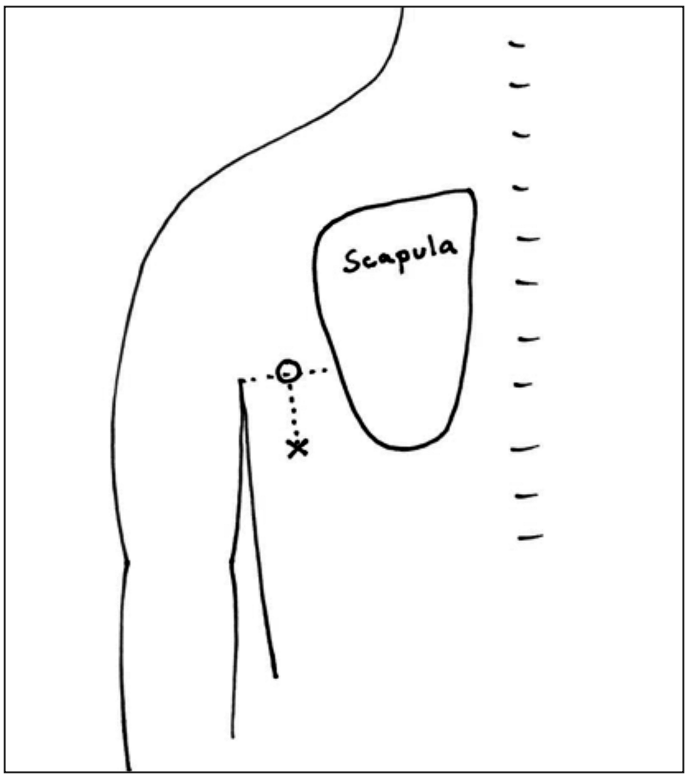

Figure 2 - The axillary measurement site was defined at a spot 5 cm below (caudal) from a line drawn between the highest point of the fold between arm and body, and the lateral scapula edge.

Table 1 - Patient Characteristics (n = 65)

Age (mean± SD)	61.2 ± 8.1 years
BMI (mean± SD)	27.3 ± 4.4 kg/m^2
Tumor size *(mean± SD)	14.3 ± 7.3 mm
Affected site n (%)	
Left	28 (43.1%)
Right	37 (56.9%)
Chemotherapy n (%)	
Yes	10 (15.4%)
No	55 (84.6%)
Surgery type n (%)	
Lumpectomy + SLNB	55 (84.6%)
Lumpectomy +ALND	10 (15.4%)
Total dose of radiotherapy n (%)	
42.5 Gy	55 (84.6%)
50 Gy	9 (13.8%)
66 Gy**	1 (1.6%)

(*) With multifocal tumors in 4 patients the size of each tumor were added up to a total sum.

(**) One patient had an extra boost of 16 Gy to the operation area.

Temporal changes of TDC

Breast

At all measurement time-points there was a significantly higher TDC value in the treated breast compared with the healthy breast (Table 2).

Figure 3 shows the mean breast TDC ratios between treated and contralateral sites at 10 different measurement occasions. There were significant differences between these mean TDC ratios ($p < 0.001$). The mean TDC ratios were indicative for edema (TDC ≥ 1.40) during the third week of radiotherapy and also at three (1.56 ± 0.04) and six months (1.50 ± 0.03) post-RT. The mean TDC ratio was 1.27 ± 0.03 at base-line, and 1.23 ± 0.03 at two years after RT treatment. The highest ratios were 1.56 ± 0.04 and 1.50 ± 0.03 which were seen at three respective six months after the treatment.

There were no differences between the mean TDC ratios during RT treatment, shortly (at two and four weeks after RT) and at one year after the treatment. Significant differences were found between the mean TDC ratios before RT and other time-points ($p \leq 0.02$), between the mean TDC ratios at three and six months after RT and other time-points ($p \leq 0.001$), and between the mean TDC ratios at two years after RT and other time-points ($p \leq 0.02$).

The proportions of patients exceeding the edema threshold limit (TDC ratios ≥ 1.40) at the 10 different time-points were 29%, 42%, 40%, 43%, 39%, 40%, 63%, 63%, 39% and 28%.
The largest proportion of patients with TDC ratios were found at three and six months post-RT and the smallest proportions at two years post-RT.

Axilla and arm

TDC measurement at axilla and arm were following: at baseline, the average TDC values were 22.4 ± 4.6 and 22.1 ± 3.8 for the operated and healthy arm sites, and 30.1 ± 4.7 and 29.1 ± 4.5 for the operated and healthy axillary sites. No statistical significant changes between treated and non-treated side were detected across the 2 years follow-up.

Temporal changes in TDC between different patient groups

The breast TDC ratios in patients in the SLNB or ALND group were not statistically different at each measurement time-point showing no difference in increase of edema in the breasts in these subgroups of the patients. Differences between proportions were neither found in the subgroups of patients having edema indicating or non-edema indicating breast TDC ratios. At three months after RT there was a tendency towards a difference in edema indicating or non-edema indicating breast TDC ratios across these subgroups ($p = 0.08$, Table 3).
There were no significant differences between these subgroups concerning TDC values on both axilla and arm at each time-point.
b) No significant differences were found concerning breast/ axilla/ arm TDC ratio between patients given hypo-fractionated or standard fractionated radiotherapy.

BMI

Before RT, 49,2% of the patients had overweight (BMI>25 kg/ m^2) and 24,1% were obese (BMI>30 kg/ m^2), with no significant change during the follow-up period.

Table 2 - Comparison between TDC values (mean ± SD) in the treated and healthy breast at 10 different time-points

Time-point	Operated breast	Healthy breast	p-value*
Before start of RT	34.8 ± 8.5	27.5 ± 5.0	$1.9*10^{-8}$
After first week of RT	37.9 ± 9.1	27.9 ± 4.8	$1.2*10^{-12}$
After second week of RT	38.1 ± 8.7	28.4 ± 6.4	$5.4*10^{-11}$
After third week of RT	38.9 ± 9.4	28.0 ± 4.4	$5.0*10^{-11}$
Two weeks after RT	38.0 ± 9.1	28.8 ± 5.1	$8.3*10^{-11}$
Four weeks after RT	38.5 ± 8.5	28.8 ± 4.6	$3.4*10^{-13}$
Three months after RT	44.4 ± 8.6	28.6 ± 3.8	$<1.0*10^{-18}$
Six months after RT	42.6 ± 7.5	28.7 ± 4.0	$<1.0*10^{-18}$
One year after RT	37.0 ± 7.8	27.6 ± 3.5	$5.8*10^{-15}$
Two years after RT	32.8 ± 6.2	26.8 ± 3.4	$3.1*10^{-10}$

(*) A value $p < 0.05$ was chosen as significance level.

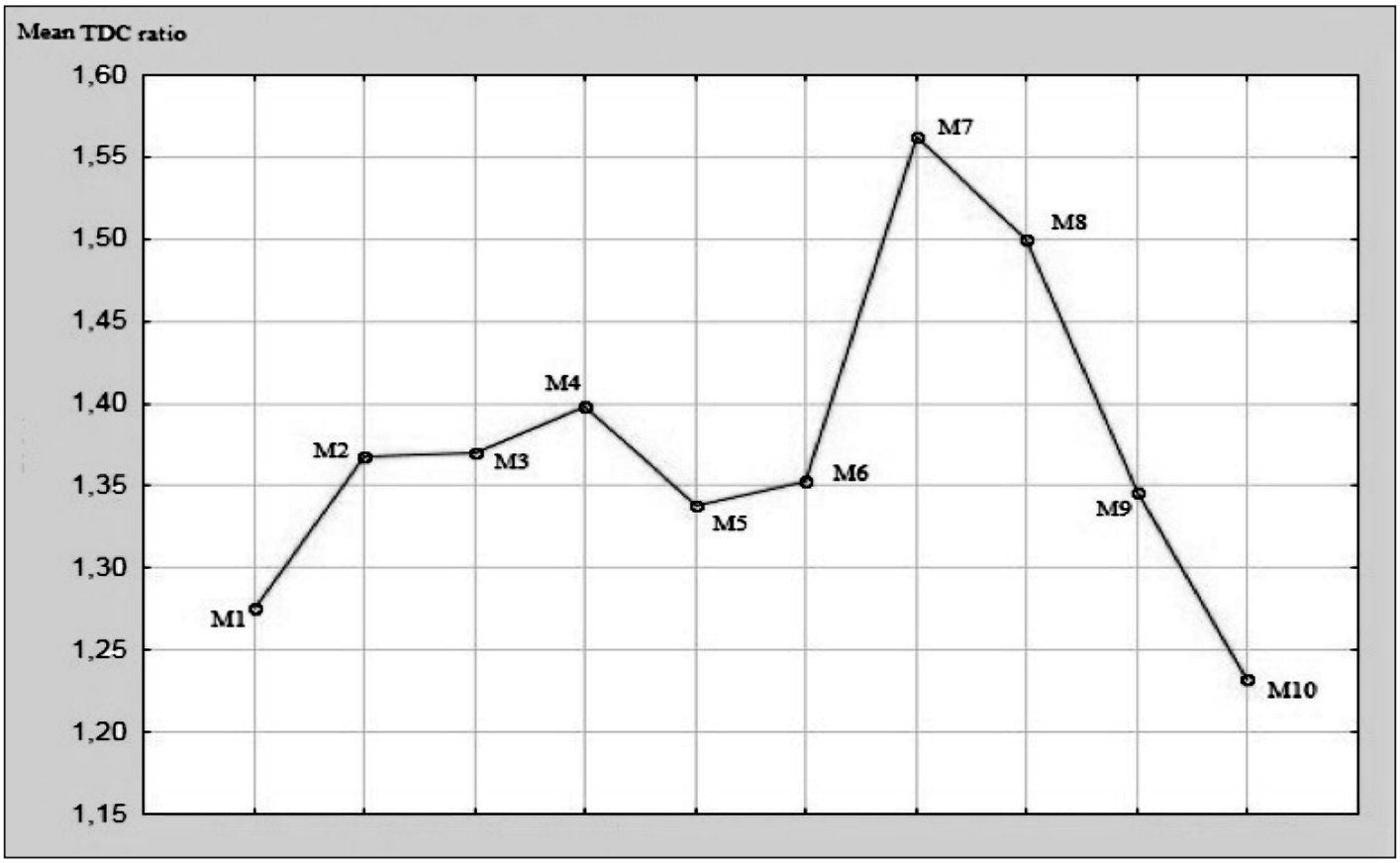

Figure 3 - Graphical comparison between mean breast TDC ratios at 10 different time-points Note: M1=at start of RT, M2, M3, M4=time-points during RT, M5 and M6=two respective four weeks after RT M7, M8, M9 and M10=three months, six months, one year respective two years after RT.

Table 3 - Proportion of patients with TDC ratios indicative for breast edema (TDC ≥ 1.40) and without significant breast edema (TDC < 1.40)

Patient group	TDC ratio ≥1.40		TDC ratio <1.40		Two-tailed p-value*
Time-point	SLNB	ALND	SLNB	ALND	
Before start of RT	15 (23.1%)	4 (6.2%)	40 (61.5%)	6 (9.2%)	46
After first week of RT	23 (35.4%)	4 (6.2%)	32 (49.2%)	6 (9.2%)	100
After second week of RT	23 (35.4%)	3 (4.6%)	32 (49.2%)	7 (10.8%)	73
After third week of RT	24 (36.9%)	4 (6.2%)	31 (47.7%)	6 (9.2%)	100
Two weeks after RT	22 (33.8%)	3 (4.6%)	33 (50.8%)	7 (10.8%)	73
Four weeks after RT	23 (35.4%)	3 (4.6%)	32 (49.2%)	7 (10.8%)	73
Three months after RT	32 (50.0%)	9 (14.1%)	22 (34.4%)	1 (1.5%)	8
Six months after RT	33 (50.8%)	8 (12.5%)	22 (33.8%)	2 (3.1%)	30
One year after RT	20 (30.8%)	5 (7.7%)	35 (53.8%)	5 (7.7%)	49
Two years after RT	14 (21.5%)	4 (6.2%)	41 (63.1%)	6 (9.2%)	44

(*) The comparisons were done with using Fisher's exact test.

DISCUSSION

In the present investigation we examined tissue water content in skin and upper subcutis in women treated for breast cancer with breast conserving surgery and RT to the breast, in order to provide information about development of edema, during the two years post-treatment.
Results demonstrated that absolute TDC values in the treated breasts were significantly greater than in the healthy breasts at all measurement time points.
The results further showed that the mean TDC ratio increased slightly during the radiation treatment but gradually diminished over the course of the next four weeks.
The TDC ratio increased again reaching the greatest value at three months after radiation treatment. The mean TDC ratio decreased again after this time-point, reaching the lowest value at two years. The temporal pattern showed that at 24 months post-RT, 28% of patients demonstrated a breast TDC ratio equal or greater than the 1.40 edema threshold limit.
The high incidence of breast edema at two years after the treatment, and also the nonlinear and wavy pattern of TDC ratio (Figure 1) during this follow-up period could reflect occurrence of edema of different origin. For example a radiation induced edema occurring several weeks (2-4 weeks) after completion of RT with an induction of inflammatory markers which may change the vascular permeability of breast tissue, and breast edema becomes a reality [9]. In opposite to another delayed radiation reaction at 3 to 6 months post-RT caused by the killing of vascular endothelial cells blocking the vessel lumen and resulting in plasma leakage to extravascular space.
Differences in surgical procedure impact the incidence of arm lymphedema. According to McLaughlin et al. [10] and Mansel et al. [11] the incidence at one year was 13% and 19%, for patients undergoing ALND compared to 5% and 3% for patients with SLNB, respectively. Moreover, the incidence of ipsilateral arm edema is clearly associated with both the extent of axillary surgery and radiation therapy [12].
In the present study, comparison of the breast, axillary or arm TDC ratios or the proportions of patients with increased breast TDC ratios for the different groups of patients showed no significant differences between these groups at any measurement occasions (Table 3). This might be due to a small number of patients with ALNB and small number of patients with higher radiation dose.
Despite the insignificant changes in body weight during two year follow-up period, the TDC ratio between treated and healthy breast changed significantly after the radiation treatment. Therefore, BMI cannot be confirmed as a factor affecting the TDC ratios between affected and contralateral side. This finding is in agreement with previous results from breast cancer related arm lymphedema [13,14].

CONCLUSION

Many patients have breast edema following breast cancer therapy. Based on the present findings, 29% had this complication already before radiation treatment. A very frequent transient edema occurs at three to six months followed by decrease of the incidence of edema after one and two years. Although the number of observations was small, differences in the surgical procedure in SLNB and ALND are unlikely to change the incidence of breast edema or lymphedema in axilla and upper arm during a two-year follow-up period.

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PRESSOTHERAPY: INTERFACIAL PRESSURE ALWAYS IN EXCESS?

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ABSTRACT

Introduction

The main problem optimizing the conservative treatment of a resistive lymphœdema is the choice of the effective compression. What has complicated the problem is that the compression value of a pneumatic pressotherapy has been presented to be in excess between cuff and the skin. This has led to block the therapeutic pressure to low value, disregarding the compliance of the support on which the pressure is exerted.

Objective

Does the interfacial pressure varies with the compliance of the compressed element?

Method

The impact of intra-cuff pressure on the interfacial ones has been evaluated by a Kikuhime®, TT Meditrade™ , Denmark during a 40 mmHg cuff inflation on three cylinders that had the same diameter but a different tonometric compliance: 0, 1.1, 2.3 mm.

Results

By modifying the compliance of the compressed element, it appeared that on a firm element (tonometric compliance of 1.1 mm) the intra-alveolar pressure equalizes almost that measured in interface: 40 mm Hg = ± 40 mm Hg. The interfacial pressure increased (25-45%) to 50-56 mm Hg on a solid cylinder (tonometric compliance of 0 mm). It decreased (-10 to -15%) to 36-34 mm Hg on a softer cylinder (tonometric compliance of 2.3 mm).

Conclusions

It is random to choose the value of compression of a lymphœdema on basis of the fear of an intra-alveolar pressure excess. Let's talk the talk! External pressure does not always produce an excess on the skin. The compliance of the element compressed seems a dimension of very first order in the interfacial pressure and thus in an effective protocol of decongestion.

Key words: compliance, interfacial pressure, lymphœdema, pressotherapy, tonometry.

INTRODUCTION

The interface pressure (IP) is a, more and more, used way for the education and the training of bandage setting/stocking class, pressotherapy, and massages. It is an easy and a speedy way to quantify the pressure by a therapeutic medium on the skin too. But, to hold the interface pressure to be a test of reference has to be viewed cautiously. Effectively and surprisingly, results did have a very low reproducibility [1]. Even though the sensor is placed in the same geometric shape of the limb segment on a same radius curvature and compressed by the same external pressure, the same external/interface pressure values are not found … over swelling. Except over bony prominences, standard deviation may be much higher than 10%. The Laplace transform cannot explain such differences and variations.
This has to be explained by other factors; i.e. the magnitude of the pressure transmitted into the skin. This phenomenon depends on several factors - surface (Pascal's law - 1647) /duration of compression, thickness of tissue compressed - inter alia the compressibility or viscoelasticity of tissues; especially in pathological conditions as swelling/fibrosis [2]. In addition, the magnitude of the pressure transmitted will changes with the swelling displacement that depends on plasticity and deformability of its environment (Coulomb's law - 1781) and the residual microcirculatory resorption function.
Closed cylinders will allow neutralizing this last factor. For the factor of compressibility, the wall of such cylinders must therefore have the property of more or less deformability under the effect of compression, i.e. more or less compliance. With such closed and deformable cylinders, the influence of tissue/swelling consistency on IP would be under satisfying observation.

MATERIAL AND METHODS

Three closed cylinders of a same 80 mm-outer diameter were used and placed in a horizontal position. Their compliance-value was performed by a weight-based tonometre BME 1428 (Flinders Medical Center; Adelaïde; South Australia) that measures the indent depth (in mm) under the effect of a standard vertical compression. At the mid length of the cylinders (x cm), a pneumatic cuff was folds around and fixed by a hook-and-loop fastener, i.e. a Velcro® band. It provides an evenly contact but no basal compression. The

dimensions of inflatable cuff were: 12 cm in the width; 27 cm length.
It was, successively, recorded to two sequential pneumatic pumps: a Hydroven12® with 12 compartments (Flowtron™ , England) and an *i-Press*1.0® with 7 compartments (Electronics of Mazet™ , France). The selected pressure of 40 mm Hg was pre-set on each pump.
Cuff/tissue IP was measured with a Kikuhime® (TT Meditrade™ , Denmark) device. This system consists of a thin (0.3 cm in its middle) circular (diameter: 3.5 cm) air cell sensor. Between the cuff and the compressed surface, the middle of its ellipsoidal thickness was fixed to the cuff by a double-side adhesive, just facing the middle of the inflatable portion of the cuff. IP data were read on its digital dial and noted in mm Hg.

RESULTS

When there is no tonometric indent depth (0 mm) (Table 1), an increase of IP appears lesser important with the Lymphassist® (50 mm Hg; +25%) than with the i-Press® (67 mm Hg; 67.5%). Over a rigid plastic (tonometric indent depth of 1.1 mm) the intra-alveolar pressure (40 mmHg) equalizes almost that measured in interface: 39 mm Hg (–2.5%) with the Lymphassist®; 42 mm Hg (+5%) with the i-Press®. Finally, over a semi-rigid plastic (2.3 mm tonometric indent depth), a decrease (of respectively –15 to –10%) is noted (34 vs 36 mm).

Table 1: ***Interface pressure variations under a cuff inflated at 40 mm Hg on three cylinder model that have different compliance by two sequential pneumatic pumps.***

CYLINDER TYPE	Tonometry indent depth (mm)	Intra-cuff pressure (ICP) (mm Hg)	INTERFACE PRESSURE (IP) in mm Hg (%óIC/IP)	
			Lymphassist®	i-Press®
Glass	0	**40**	50 (+25)	67 (+67.5)
Rigid plastic	1,11		39 (–2.5)	42 (+5)
Semi-rigid plastic	2,3		34 (–15)	36 (–10)

DISCUSSION

Firstly, when tonometry indent depth is of 1.1 mm, the IP is near the value of the IC (39 or 42 mm Hg vs 40 mmHg). But in general, the IP is not equal even though the radius of the curvature – to which 40 mm Hg are applied – is equal.
The deformability of the compressed surface was shown to alter the value of the external pressure on the IP. Thus simple tonometer of measuring indent depth may allow us to develop a greater understanding of the properties of the dermis/swelling, which may have therapeutic implications like the pressure to exert on an edema [3, 4]. However, there is no nomogram to illustrate the relationship between the IP and the deformation characteristics of compressed tissue. There is no consensus too about the usefulness of tonometer measurements.
More, the tonometer does not give a real compliance value (form.1). It does not give the value of compression and the size of the compressed volume. It only gives an approach of a local tissue resistance to compression. It only measures an instantaneous depth that its plunger penetrates into the tissue. This plunger is made for flat surface and not for ellipsoidal/circular one. Its application is limited to a vertical use [5]. The small dial is difficult to read. For those reasons, this device is not currently available anymore. The coming viscoelastic indurometer (Flinders Medical Center; Adelaïde; South Australia) would be of more interest.

Formula 1: Compliance (C) is defined as the change (∂) in volume (V) brought about by a unit change in pressure (P). Compliance can be expressed as: C = ∂ V / (V x ∂ P)

A second observation: the IP magnitudes produced by the two devices differed considerably [5-42.5% ó]. That is surprisingly because of the two pumps tested are equipped with an intra-alveolar pressure controller that is made to close the valve when the pre-setting pressure is reached. We may suspect the different contact area of their respective pneumatic compartments. But in order to circumvent this potential bias the two pumps were recorded to a same cuff. So, we may suspect that their pressure controller would be made with different standards.
Third, the present results confirm that an IP may be higher than the ICP. In accordance with the Laplace's law, it is known that IP is a function of the ICP and the cylinder radius/geometry. Thus, because of the smaller cylinder radius – compared to the one of the cuff – it would be normal to find an IP higher than the IC one. But because of a same radius of the 3 cylinders, there must be no different response between their IP.
This over-estimation was lesser (+25%) with the Lymphassist® than with the i-Press® (+67.5%). An over-estimation of 80% has been reported in the literature that was considered as excessive [1]. Maybe the suspicion of such relative "excessive" pressure do have lead the conceptors of the Lymphassist® program to restrain the inflation to a lower value than that indicated by the pump manometer.
The qualification of "excessive" is somewhat excessive for three main reasons:

1. an oedema is not a pressure but a volume in excess. So whether it is excessive or not, it is the effective pressure to reduce the amount of oedema that matters, not its value.
2. an "excessive" compression has no sense and is to differentiate to a non- or contra-productive pressure. A contra productive pressure that predispose to a risk of side effects depends of several factors: pressure value, shear, radius of curvature, duration, temperature, regional humidity, plasticity of the skin (fibrosis), oedema compliance variations, tolerance of the tissues before damage may occur, underlying disease as diabetus, arteriopathy, insensibility, their interactions [2,6].
3. the IP over-estimation only occur when it is applied on a hard surface that acts as a multiplier. However in the case of a consistent oedema limed in a fibrotic tissue, the external pressure rapidly dissipates in the depth. The pressure loss is proportionate to the energy transferred in the tissues. Thus, the external pressure must be raised over 100 mm Hg high to only obtain 30 mm Hg in consistent subcutis [7].

Fourth, opposite to this IP over-estimation, there is a consequent IP loss over a lesser compliant cylinder that is known to have better P/distributing capabilities. With a tonometry indent depth of 2.3 mm, the IP reduction was limited to only 10-15% of the IC (40

mm Hg). This is, maybe, the result of a greater penetration that leads to more contact to a greater surface and to a subsequent better P distribution.

CONCLUSIONS

Under the same light compression force (IC = 40 mm Hg) on a same radius of curvature, the IP magnitudes depend on the compliance of the compressed area and on the device. We found a discrepancy between the ICP and the IP that is due to the consistency of the media compressed. More the rigidity of the cylinder is high; more the IP is over-estimated. Otherwise, of a little rigidity, take a little under-estimated IP.
According to those results, consider the P of interface as a repository is very simplistic. All environmental forces (intra-pump, compressed tissue nature, etc.) interact more or less significantly. These interactions can change not only the radius of curvature but also the geometric shape of the limb segment compressed.
So it is random to choose the compression value of a lymphœdema on basis of the fear of an ICP excess. Let's talk the talk! External pressure does not always produce an excess on the skin. The compliance of the element compressed seems a dimension of very first order in an effective protocol of decongestion and there is still to learn from Lymphedema before you can determine the optimal external pressure to drain it.

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PATIENT EDUCATION PROGRAM: SCHOOL OF LYMPHEDEMA PREVENTION

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ABSTRACT

Introduction: Patients who undergo lymphadenectomy need advice on preventive care to avoid the development of lymphedema [(1)].

Objective: To present our patient educational program, the School of Lymphedema Prevention after Breast cancer Surgery.

Material and Methods: Prospective observational study of the rehabilitation outpatient medical consultation assessed woman. Patients were referred from Department of Gynecologic Oncology, Oncology and Radiotherapy [(1)].
Preventive measures were explained individually in the first appointment, prevention standards and home exercises were delivered. If they required lymphatic drainage therapy, they were recruited for the School of Lymphedema Prevention. A monthly meeting was organized up to 14 people. It was opened to patients and one family member was allowed. We informed about what the lymphedema is, how to prevent and treat it.
Then, we established a roundtable where patients could express their worries and concerns. At the end of each session, a satisfaction survey was given. Material Resources: meeting room, computer, projector and roundtable. Human resources: rehabilitation physician and skilled physiotherapist.

Results: A total of 27 meetings were conducted from 17 March 2010 until 20 March 2013 and 184 people attended to School of Lymphedema Prevention. The survey showed high degree of satisfaction.

Conclusions: School of Lymphedema Prevention focuses on training and educating patients at risk of developing lymphedema or suffering from lymphedema and its families. Emphasis is on prevention, promotion of autonomy and activities of daily living, psychological and emotional support and reducing waiting list.

Key Words: lymphedema, prevention, rehabilitation, mastectomy, school, education.

INTRODUCTION

Lymphedema is a chronic swelling of a body part and often the unintended consequence of cancer surgery and/or radiation [(1)].
Since the introduction of Combined Decongestive Therapy, the medical community has been capable of offering effective treatment. Lymphedema therapists and educators have realized about the importance of patient education in the prevention and management of this condition. Unfortunately, as there is no "cure", the long term maintenance inevitably becomes the responsibility of the patient [(2)].
The aim of creating the School of postmastectomy Lymphedema Prevention was to prevent the onset of lymphedema or to slow its progression in postmastectomized women who had underwent lymphadenectomy associated or not to radiation therapy [(1,2)].
The implementation of prevention programs allows an early analysis of individual risk of suffering from lymphedema and consequently, it is possible to design a therapeutic attitude and early treatment.
According to the reviewed literature, patients who already had lymphedema after breast cancer treatment had lower disability of the arm after attending to rehabilitation program [(3)].
In our hospital, this patient educational program was established as innovative and specifically to meet the demands and expectations of these women, providing the closest possible attention and tailored to their needs.

OBJECTIVE

Our aim is to present our patients educational program, School of Lymphedema Prevention after Breast cancer Surgery.

MATERIAL AND METHODS

It is a prospective observational study of the rehabilitation outpatient medical consultation assessed woman. Patients were referred from Department of Gynecologic Oncology, Oncology, Radiotherapy and others.
Preventive measures were explained individually in the first appointment, prevention standards and home exercises were delivered [(4,5,6)]. If patients required lymphatic drainage therapy, they were recruited for the School of Lymphedema Prevention. A monthly meeting is organized up to 14 people. It is opened to patients and one family member is allowed. We inform about what the lymphedema is, how to prevent and treat it. Patients have the

opportunity to learn the basic anatomy and physiology of the lymphatic system, different ways and measures to prevent its appearance and also its development. So that, they have the choice to understand better the risks (trauma, burns, insect bites, compression, and local infections), their own condition and also the measures to implement self-care techniques [7]. Then, we establish a roundtable where patients can express their worries and concerns. At the end of each session, they are given a satisfaction survey. The survey is divided into four sections. Initially, we question the content of the talk and it is scored by 3 degrees (absolutely, not really, absolutely not). After, we ask about the place, schedule and talk time also scoring it in 3 ranges (very suitable, suitable, and not very suitable). Then, we evaluate the overall satisfaction of participants in very interesting, interesting, not very interesting. Finally, an assessment is requested from 0 to 10, with 0 being the worst possible score and 10 the highest. (Table 1)

Material Resources: meeting room, computer, projector and roundtable.
Human Resources: rehabilitation physician and skilled physiotherapist.

Table 1

	Absolutely	Not really	Absolutely not
Do you think what you learned in the workshop will help you to prevent the development of lymphedema?			
Do you think what you learned in the workshop will help you to take care of yourself?			
Do you believe that your self-esteem, self assessment and view of your self are better after the workshop?			
	Very suitable	**Suitable**	**Not very suitable**
What do you think about the place where the workshop took place?			
What do you think about the schedule?			
What do you think about the length of the workshop?			
	Very interesting	**Interesting**	**Not very interesting**
In general terms, what do you think about the workshop?			
Rate your satisfaction level (0-10)?			

Legend: This is the survey which was given to all the patients and relatives who assisted to the workshop. It is divided into four sections: about the content, the organization (the room, schedule and length) the overall satisfaction and satisfaction level from 0 to 10, with 0 being the worst possible score and 10 the highest.

RESULTS

A total of 27 meetings were conducted from 17 March 2010 until 20 March 2013, 184 people attended to School of Lymphedema Prevention from which 20 patients did not answer to the survey and were excluded for the results.
The survey showed high interest in the content: 95.7% thought what learned was helpful to prevent development of lymphedema, to take care of them-selves. They agreed in the schedule and length of the workshop but unfortunately 10.3% found not very suitable the place where the talk took place. 84.1% showed high degree of satisfaction in general terms and the average rate was more than 9. (Table 2)
Subjectively, they were more willing to make the commitment, to learn and to implement self-care techniques.

Table 2

	Absolutely	Not really	Absolutely not
Do you think what you learned in the workshop will help you to prevent the development of lymphedema?	157 patients, 95.7%	7 patients, 4.3%	0 patients
Do you think what you learned in the workshop will help you to take care of yourself?	156 patients, 95.1%	5 patients, 3%	3 patients, 1.9%
Do you believe that your self-esteem, self assessment and view of your self are better after the workshop?	101 patients, 61.5%	63 patients, 38.5%	0 patients
	Very suitable	**Suitable**	**Not very suitable**
What do you think about the place where the workshop took place?	80 patients, 48.8%	67 patients, 40.9%	17 patients, 10.3%
What do you think about the schedule?	103 patients, 62.8%	59 patients, 36%	2 patients, 1.2%
What do you think about the length of the workshop?	105 patients, 64%	57 patients, 34.8%	2 patients, 1.2%
	Very interesting	**Interesting**	**Not very interesting**
In general terms, what do you think about the workshop?	138 patients, 84.1%	26 patients, 15.9%%	0 patients
Rate your satisfaction level (0-10)	10 points: 90 patients, 54.9% 9 points: 54 patients, 32.9% 8 points: 11 patients, 6.7% 7 points: 9 patients, 5.5% 6 or less points: 0 patients		

Legend: A total of 145 patients from 184 answered to the survey.

DISCUSSION

Secondary lymphedema after surgery for breast cancer that involved dissection of axillary lymph nodes is a complication of the patients [8]. In order to this, emphasis should be placed on prevention. Early physiotherapy with educational strategy was associated with a lower risk [3]. Nonetheless, most of the studies showed that women were not adequately informed after surgery and/or radiation therapy.

Our team has implemented an educational strategy for the patients at risk of developing lymphedema and also for those who have already developed. Our aim was to improve care quality, avoid further complications and to reduce more individualized physiotherapy treatments, so that we could manage in a better way our public resources.

We believe that our study shows evidence of high degree of satisfaction and commitment for using self-care measures. Unfortunately, there is no data of the decrease development of secondary lymphedema due to different particular criterion for diagnosing a measuring lymphedema. In other hand, the fact of including patient's relatives could increase the participation in the program as a feed-back or external support.

Nowadays, we are developing a more complete educational program which will include combined aerobic, stretching and resistance training exercises.

CONCLUSIONS

The School of Lymphedema Prevention focuses on training and educating patients at risk of developing lymphedema or suffering from lymphedema and its families. Emphasis is on prevention, promotion of autonomy and activities of daily living, psychological and emotional support.

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AXILLARY WEB SYNDROME AFTER SENTINEL LYMPH NODE BIOPSY (SLNB)
ASSESSMENT AND INCIDENCE

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ABSTRACT

Objective: Assess the incidence of Axillary Web Syndrome (AWS) or Cording in women patients who were subject to breast conserving surgery (quadrantectomy) and sentinel lymph node biopsy (SLNB). The outcome was assessed to determine degree and identify connections with other local outcomes as well as postural and emotional attitudes.

Materials and methods: 164 women subject to quadrantectomy with SLNB and no axillary dissection were observed. They were selected following the order of admission to the operating room with a single surgery team. The patients were checked before surgery, immediately after surgery and after 3 and 6 months.

Results: Through a more refined semiotics and a dedicated follow-up, AWS incidence rate turned out to be significant (33%) and the serious degree of the results persistent over time. The reported incidence rate is higher than that referred to in breast and physiatry literature, since our statistics also include subclinical AWS.

Conclusions: Women with AWS showed a high rate of sensory-motor alterations that occur prior to surgery and are related to the emotional diagnostic trauma. The high AWS incidence rate and the motion/emotion connection calls for preventive treatment of patients through combined therapies.

Key Words: AWS, Iatrogenic outcomes after quadrantectomy and sentinel lymph node biopsy (SLNB), Motion and E-motion.

INTRODUCTION

According to breast specialists, iatrogenic injuries in women who have been diagnosed breast cancer and undergo quadrantectomy with sentinel lymph node biopsy are very scarce or non-existent and there is no need to set protocols for prevention and treatment of outcomes.
Consequence thereof is a post-operative discomfort in women, which is ignored by clinicians, although ironically great attention and professional skills are currently dedicated to breast cancer treatment and no study protocols are set to prevent and treat early and late postoperative outcomes.
What goes unreported is that cancer diagnosis makes women vulnerable and that their body differs in posture, structure, constitution, physical and metabolic condition, as well as in emotions and relationship with the world after cancer diagnosis [1, 2, 3, 4].
A biomechanical and emotional investigation is therefore required before surgery.

Based on our studies on physicality and relations [5] in women who have been diagnosed with breast cancer, more than half of them unknowingly express the emotional trauma through a sensory-motor alteration consisting in hypo-pendularism of the ipsilateral arm while walking.
In the present study all the patients who were about to enter the operating room for conservative breast and axillary surgery were observed in clinical, biomechanical and emotional terms, to see if the different metabolic, structural, postural and emotional states can affect occurrence rate and degree of postoperative outcomes.
The physical outcomes that are inevitably due to the surgical incisions of the skin, the fascia and the glands – even of slight degree – add up with and are affected by these preceding and different "physical" features, which often are not encoded in protocol studies on iatrogenic injuries.
Purpose of this work – carried out in a breast surgery unit in the process of accreditation at Breast Unit – is to assess the degree of a specific outcome that impairs the arm movement in relation to the world, an outcome generally referred to as Axillary Web Syndrome (AWS) or Cording, by connecting it to women's e-motion after diagnosis.

AWS O CORDING

Incision of the skin in the chest and underarm region, opening of the clavipectoral fascia with sentinel lymph node biopsy (SLNB) may lead to post-operative development of either one or several smaller or thicker cords, that extend down from the site of scarring to the inner arm and forearm and sometimes continue all the way down to the wrist.
These cords lead to motion restrictions, feeling of tightness and pain. The morphology of the cords with their different pathways, tightness and size, makes etiopathogenetic interpretation difficult. Some authors talk of sclerosis of lymphatic vessels and veins with surrounding fibrosis.
Such manifold picture is clinically defined as retraction, adhesion, lymphatic sclerosis and fibrosclerosis [6]. For a decade, this symptom has been known as "Axillary Web Syndrome (AWS) or Cording" [7, 8].
Based on literature data, AWS occurs in 20% of women who have been subject to SLNB. A great number of breast specialists consider this rate overstated, as during post-surgical visits they rarely find a picture of AWS or Cording.

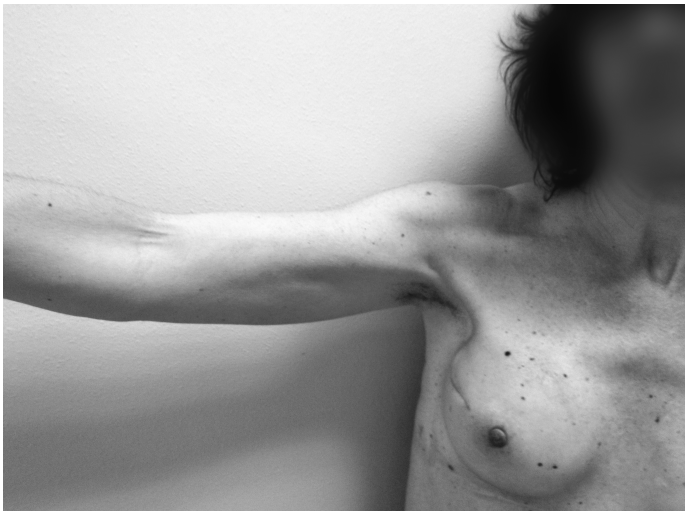
On the other hand, literature rarely refers to the immediate complications, to the extent of glandular removal and incisions of the fascia, to the correlations with the conditions of the tissues, the fascia, the dynamics of the shoulder, the musculoskeletal and metabolic structure and sensory-motor changes due to diagnosis trauma.

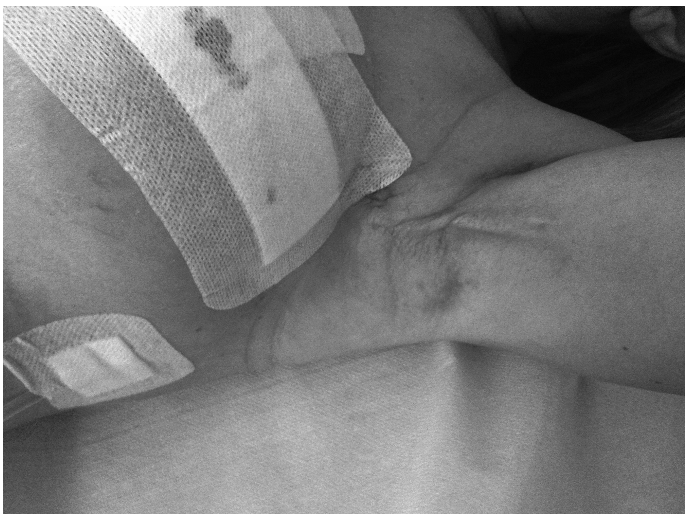
MATERIALS AND METHODS

The study assessed the incidence of AWS after qudrantectomy and SLNB based on new parameters and tried to understand the incidence of both the other post-surgical complications and the emotional, structural and functional conditions at disclosure of diagnosis in the group of women observed.
Between the beginning of 2012 and the end of 2013, a sample of 164 women was selected; they had undergone quadrantectomy and SLNB surgery consecutively and with a single surgical team following standard procedures [9, 10, 11, 12]. Patients were checked prior to surgery, after 10, 20, 30 days and 2, 3, 6 months.
Assessment of the AWS picture was rated according to subclinical parameters (Grade G 0, where cording is reflected by the skin sliding from the shoulder towards the elbow with no feeling of tightness and impairment) and clinical degrees (G1, G2, G3).
G1 includes small superficial cords up to the elbow with slightly limited range of motion; G2 refers to one or several thick cords that extend down to the elbow and also impair elbow extension; G3 refers to cords that run down to the wrist with limited range of motion of elbow and wrist.

RESULTS

The study that was carried out on 164 patients undergoing consecutive quadrantectomy surgery with SLNB without axillary dissection, by our breast surgery team, resulted in low incidence of known complications (serosity, infections, impairments) and high AWS incidence.
Average age in the group of patients was 54.2. The percentage of overweight women was comparable to the average of Italian women, not over 31%; 2% were obese, 67% were in menopause, 25% had hypertension and metabolic diseases controlled with medications, 10% moderate depression.
Only 3% of women had shown excessive serosity that required drainage for over 15 days and 1.8% had had to extend antibiotic therapy for local low-intensity infections. The cords, either thin or thick, differing in paths, both painful and impairing, occurred in 54 patients (33%).
The high AWS percentage in the group of women who were operated in our breast unit is due to the fact that our rating considers a subclinical grade 0, where cording is reflected by the skin sliding from the shoulder towards the elbow with no feeling of tightness or limitation.
G1 includes small superficial cords up to the elbow with slightly limited range of motion, G2 refers to one or several thick cords that extend down to the elbow and also impair elbow extension; G3 refers to cords that run down to the wrist with limited range of motion of elbow and wrist.
This percentage is higher than that referred to in literature, simply because AWS has been better observed and coded. In the group of women showing visible cords that impaired the extension of the arm (43 patients with G 1, 2, 3), 11 cases were subclinical Grade G-0, only visible with sliding movement.
The group of patients with AWS showed an increased incidence of sensory-motor changes with hypopendularism and haptic hypofunction of the hand (tactile and motor exploration), assessed after diagnosis at the biomechanical investigation before surgery, compared to patients who had not developed AWS (68.8% vs. 35%).





Women with AWS entered a randomized study on the efficacy of lymphokinetic, anti-inflammatory and mood- modulating remedies. The control group was free to take or not take self- and hetero-prescribed medications, depending on the symptoms (pain, limitation, inflammation, anxiety).
After two months, 70% of G2 and G3 cording was still present.
Half of the women were to follow a cyclic motor and manual rehabilitation therapy and constant clinical follow-up. Chronicity worsened in cases of adjuvant therapies such as radiotherapy and chemotherapy.

Stretching and rupture of the cords to reduce tension in the arm extension were never prescribed, since in our experience, besides the pain complained by the patients during the breaking stage, there had been repercussions in fibrotic networks as well as transient lymphatic stasis.
The gentle approach of manual lymph drainage proved more effective compared to the traumatic manual rupture of the cords. Rehab therapists provided for dissection of the scar, mobilization of the clavicle, shoulder and arm as well as laser therapy and taping [13].
A psychomotor and psychological approach by dedicated therapists was required for those patients showing a significant sensory-motor weakness, as well as very low emotional and relational haptic features.

CONCLUSIONS

SLNB, which is considered a low invasive and side effect-free technique, often results in outcomes that might escape regular breast screening. For instance, AWS incidence rate can be high if you carefully consider tightness and subclinical retractions that can lead to an increase in reactivity of fibroblast adhesion and reticular fibers.
Fascial tightness before surgery and sensory-motor alterations caused by the trauma of diagnosis, drive us towards treatments that take into consideration both connective and emotional reactivity. Persistence of grade 2 and 3 conditions after two months from onset and potential worsening due to post-surgery adjuvant radio and chemotherapies should foster implementation of proper preventive pharmacological and rehabilitation protocols.

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A RARE CLINICAL MANIFESTATION OF CONGENITAL LYMPHATIC DYSPLASIA: THE EFFICACY OF COMPLEX DECONGESTIVE THERAPY IN A YOUNG CHILD

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Letter to editor

Dear Sir,

We report a child of congenital lymphedema presenting with lymphedema of right face, bilateral arms and with severe intestinal lymphectasia. A sixteen-month-old female child was consulted to our department of physical and rehabilitation medicine for the swelling of right face, arms (more noticeable in the right side) and abdomen (Figure 1). She had edema from birth on her right and left hands and arms, right side of face and diffuse edema in abdominal region. Her complaints had increased for the last four months and she had diarrhea for a long time. Her family's past medical history was unremarkable. In her physical examination she had distended abdomen and tachpneic respiration besides. Her diffuse edema was non pitting type. The scintigraphic examination was revealed as; no lymphatic drainage at the right upper extremity (severe lymphatic hypoplasia or agenesia), and delayed lymphatic drainage at the left upper extremity, and normal lymphatics at the lower extremities. These findings were concordant with primary congenital lymphatic dysplasia. Also she had ascites in the abdomen and pericardial effusion in computed tomography and echocardiography. She had no cardiac or renal failure. In her upper gastrointestinal endoscopic examination there was severe intestinal lymphectasia. She had paracentesis, pericardardial drainage and was performed total patenteral nutrition, following the proper diagnosis. After her general condition was stabilized, we started complex decongestive therapy for the lymphedema in the right upper extremity which was more prominent. Manuel lymphatic drainage and multilayered banding (with relatively low tension) techniques were applied daily, for a duration of three weeks. In addition kinesiotaping was performed for the abdominal lymphedema. In the follow up period, her measurements for edema and abdominal distention were improved gradually (Figure 2a and 2b). In this aytpical and very rare case we aimed to report the short term relative efficacy of complex decongestive therapy in such a young child. The family was educated for the risk reduction methods for complications of lymphedema and manual lymphatic drainage as well as bandaging. They were also instructed for follow-up visits. A pressure garment was planned for the control visit, 6 months later.

Most forms of primary lymphedema are thougt to be caused by a congenital abnormality of the lymphatic system [(1)]. Primary lymphedema is a rare disease; prevelance ranges within 1:6000 to 1:10.000 [(2)]. It is seen more in females. The most frequent clinical

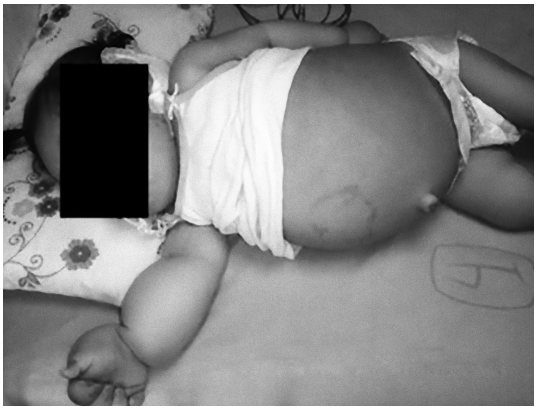

Figure 1

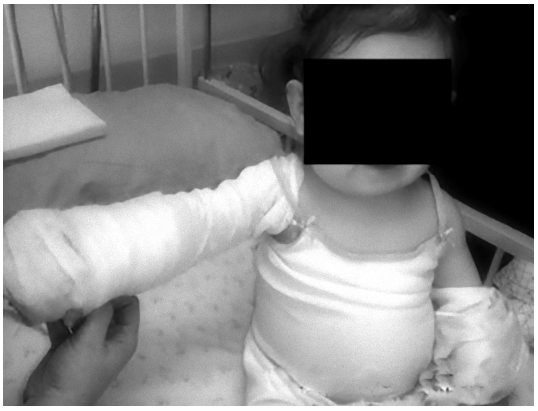

Figure 2a

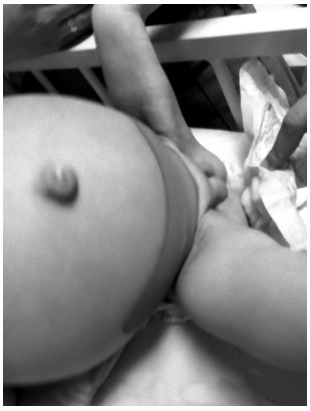

Figure 2b

presentation is swelling of the leg and ankle. However upper extremity may also be affected in congenital lymphedema [3]. Complex decongestive therapy which has individual components of skin care, manual lymph drainage, compression bandages, exercise and pressure garments, is the golden standard treatment for reducing both primary and secondary lymphedema [4].
In this case we aimed to indicate the short term effects of manual lymphatic drainage and multilayer bandaging in the right extremity. Although the regression of abdominal distention seems to be due to the parenteral nutrition with specific diet, we believe that kinesio taping may make even a little contribution.

Sincerely

Gökhan Çağlayan, MD
Specialist of PRM

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ERRATA CORRIGE

Abstract for the *41th ESL European Society of Lymphology Congress,* June 4th-6th 2015, Lausanne, Switzerland
Versione Prof. G. Pantaleo (01.04.2015)

ON THE RELIABILITY OF TONOMETRY: A PILOT STUDY OF INTER-RATER CONSISTENCY AND RELATED PSYCHOSOCIAL FACTORS UNDERLYING THE FORMULATION OF TONOMETRIC JUDGMENTS

Sandro Michelini [1], Rinaldo Caldirola [2], Simona Sciara [3], Giuseppe Pantaleo [3]

Introduction. Tonometric judgments are critical in the diagnosis and treatment of lymphedema. Yet, while some studies (e.g. Lye *et al.*, 2006) claimed the reliability of particular mechanical tonometric instruments, in summarizing the main findings of their systematic review Oremus *et al.* (2010) recently concluded that "there is too little evidence to draw conclusions about the reliability of [diagnostic] tests such as *tonometry*." (p. 4, emphasis added). **Materials and Methods.** To fill this gap – a tangible lack of knowledge of great import both for the literature and even more for good clinical practice – we instructed a team of trained raters to independently supply distinct sets of tonometric judgments with respect to (a) patients with diagnosis of lymphedema (the *clinical sample*), and (b) their healthy counterparts (the *comparison sample*). Each judgment was supplemented by the assessment of two additional distinctive features lying at the core of any judgmental process – the degree of *difficulty* and the degree of *certainty* reported by the judge in formulating tonometric judgments –. **Results and Conclusions.** We will illustrate and report on the preliminary stage of this developing research endeavor, and possibly also suggest some initial conclusions on the basis of our earliest results.

Words Count: 1300 characters (*vs.* max 2500 characters)

Keywords: tonometric judgments , tonometry, inter-rater reliability, judgmental processes

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THE INFLUENCE OF ARM SWELLING DURATION ON SHOULDER PATHOLOGY IN BREAST CANCER PATIENTS WITH LYMPHEDEMA

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The authors declare no conflict of interest.
The authors confirm that there is no financial arrangement with anyone.

Running head: shoulder pathology and lymphedema duration.

ABSTRACT

The aim of this study was to evaluate the influence of arm swelling duration on shoulder pathology in patients with lymphedema. Forty-seven breast cancer patients with unilateral lymphedema were assessed. We surveyed the duration of arm swelling and shoulder pain. The patients then participated in a shoulder ultrasound examination for lesions. We divided our patients into two groups based on a lymphedema duration of within 1 year and after 1 year. The shoulder ultrasound findings in patients with more than 1 year of lymphedema revealed that a supraspinatus tendon tear was more common. In contrast, subacromial subdeltoid bursal thickening was more common in the within 1 year group. Patients with a supraspinatus tendon tear (n = 13) revealed a significant longer duration (1310 days vs. 398 days, $P = 0.026$). Subacromial subdeltoid bursal thickening had shorter duration trend but was not significant (454 days vs. 892 days, $P = 0.058$). The duration of lymphedema influences rotator cuff tendon pathology. Clinicians should therefore adopt an early management approach to shoulder pain by obtaining a precise diagnosis in patients with breast cancer related lymphedema.

Keywords: Breast Neoplasms, Lymphedema, Shoulder Pain, Ultrasonography.

INTRODUCTION

Breast cancer related lymphedema (BCRL) and shoulder pain are common problems and considered quality of life (QOL) predictors in breast cancer patients after treatment [(1)]. BCRL can cause or aggravate shoulder pain through a decrease of motion in the affected limb, increased fluid tension in the subcutaneous tissue, an increased risk of cellulitis and other infections, lymphangiosarcoma, a decreased healing capacity in the affected tissue, and pathology in the rotator cuff tendon. In addition, shoulder pain may petentially cause lymphedema. A reduced QOL and a prolonged hospital stay can result from these complications in the breast cancer patients [(2-4)]. Hence, an accurate diagnosis and proper management of shoulder pain are necessary for reduced disability and improved QOL in patients with BCRL. However, the pathology of the shoulder pain related to BCRL has not yet been clarified. Moreover, BCRL is a chronic condition and the duration of the lymphedema may impact on the nature of the shoulder pathology.

A recent report has presented ultrasound findings for the shoulder pathology in patients with BCRL [(5)]. However, that study did not enrolled patients as BCRL duration which prevented any analysis of the cause of shoulder pain as the period after breast cancer treatment. The aim of our present study, therefore, was to assess the influence of lymphedema duration on shoulder pain in cases of BCRL through the use of ultrasound.

MATERIALS AND METHODS

Participants

This is a cross-sectional study of 47 women aged 40~78 years who had been treated for unilateral breast cancer and had subsequently developed BCRL. All participants had access to complex decongestive therapy comprised of manual lymph drainage, compression, exercises, and skin care at our lymphedema clinic. All of the patients enrolled in this study showed an arm circumference difference of > 2cm above or below the elbow [(6,7)] and the BCRL diagnosis had been confirmed using lymphangioscintigraphy of the upper limbs. All of our study subjects were free of cancer at the time of the analyses. Patients

with bilateral lymphedema, lymphangitis, skin disease, inflammatory shoulder arthritis, a previous shoulder joint trauma, or who had undergone shoulder surgery were excluded.
This study was conducted with the approval of our institutional review board (OHRP CMC OC09OISE0007). Written informed consent was obtained from all participants after they had been briefed about the study. We surveyed the time and duration of arm swelling, age, height and body weight of each patient subject. Shoulder pain in all cases was measured using a visual analog scale and each patient completed a disabilities in the arm, shoulder and hand (DASH) questionnaire.

Musculoskeletal Examinations and Ultrasound

Patients underwent a musculoskeletal examination and also an ultrasound of the affected shoulder region. The ranges of motion for the patients' shoulders were measured for flexion, abduction, external rotation, and internal rotation, using a goniometer in the supine position. Ultrasound examinations were performed using an X11 XEPhilips, Bothell, WA, USA) a 7-12 MHz line-array transducer. The physiatrist, a Korean board certified physician of musculoskeletal ultrasound medicine who conducted the ultrasound examinations was blinded to the duration of arm swelling. Each patient was seated on chair behind the examiner. In accordance with a previously described method [8], five standard ultrasound views (anterior transverse and longitudinal, lateral transverse and longitudinal, posterior transverse) were selected and a dynamic examination was also included. The subscapularis, supraspinatus, infraspinatus and biceps tendons were examined in the transverse and longitudinal planes.
The pathologic findings and diagnoses made by ultrasound were based on previously described criteria [9] as follows: a tendon tear was defined as a discontinuity of the tendon fibers; as a hypoechoic or anechoic defect; as a partial tear of the tendon fibers involving either the bursal or the articular surface that appeared as a focal hypoechoic; or as an anechoic defect. Subacromial subdeltoid bursal thickening was defined as a bursal thickness of more than 2 mm transverse with associated hypoechogenicity, with or without bursal fluid. Distention of the biceps brachii tendon sheath was defined as hypoechoic or anechoic fluid surrounding the biceps tendon. After an examination, another physiatrist, a Korean board certified physician of musculoskeletal ultrasound medicine, confirmed the findings. If there were disagreements about the interpretations, then ultrasound examinations were conducted again.
We also defined adhesive capsulitis as chronic shoulder pain and a loss of 30° or more in the passive range of motion of the glenohumeral joint in external rotation, and at least one of flexion, abduction or internal rotation.

Statistical analysis

Statistical analyses were performed with SPSS (version 16.0) software. Data are presented as mean ± standard deviation or the number (percentage) of patients. The chi-square test or Fisher's exact test and the independent sampled t-test were used in the analysis as appropriate. A P-value < 0.05 was considered statistically significant.

RESULTS

Clinical characteristics

Participants were classified into two groups according to lymphedema duration (i.e. time from first self-report of symptoms). Thirty-two patients with lymphedema had unilateral arm swelling within 1 year or less (within 1 year group) and the remaining 15 patients developed these symptoms at more than 1 year (after 1 year group). The mean age of the patients was significantly higher in the after 1 year group (57.0 ± 8.8 vs 49.4 ± 7.6). There were no significant differences between the body mass indexof the patients in the two groups. The mean duration of lymphedema was 137.6 days in the within 1 year group and 1694.3 days in the after 1 year group. We found that 50% of the within 1 year group and 86.7% of the after 1 year group cases had a history of modified radical mastectomy (Table 1). We further noted that 59.3% of the within 1 year group and 40.0% of the after 1 year group patients complained of ipsilateral shoulder pain. The visual analog scale result was significantly higher in the within 1 year groups (4.0 ± 2.1) than in the after 1 year patients (2.0 ± 0.6), but the DASH results did not show any significant differences between the two groups (Table 1).

Table 1 - Characteristics of the breast cancer patients with lymphedema analyzed in this study

Duration	Arm swelling duration ≤ 1 year (n=32)	Arm swelling duration > 1 year (n=15)	*P* value
Age (years)	49.4 ± 7.6	57.0 ± 8.8	0.006**
Body index mass (kg/m^2)	25.1 ± 3.3	24.4 ± 3.9	0.539
Type of surgery Modified radical mastectomy Breast conserving operation	 n=16 (50%) n=16 (50%)	 n=13 (86.7%) n=2 (13.3%)	0.008
Axillary lymph node dissection	n=31 (96.9%)	n=15 (100%)	0.698
Radiation Therapy	n=25 (78.1%)	n=11 (73.3%)	0.294
Duration of diagnosis (days)	137.6 ± 130.5	1694.3 ± 1885.8	0.004**
DASH	30.1 ± 16.3	35.3 ± 18.4	0.330
Ipsilateral shoulder pain	n=19 (59.3%)	n=6 (40.4%)	0.347
Visual analog scale	4.0 ± 2.1*	2.0 ± 0.6	0.006**

DASH, disabilities in the arm, shoulder and hand questionnaire. **P < 0.01.

Ultrasound findings and clinical features

Abnormalities were detected by ultrasound in 87.2% (41/47) of all study participants. Among the 41 patients in our study cohort with abnormal ultrasound findings on the arm swelling side, 27 cases were included in the within 1 year group (27/32, 84.4%) and 14 patients were assigned to the 1 after year group (14/15, 93.3%). The abnormal ultrasound findings in the shoulders of our patients are listed in Table 2. A supraspinatus tendon tear was more common in the after 1 year group (53.3% vs. 15.6%) and subacromial subdeltoid bursal thickening was more common in 20/32 (62.5%) patients in the within 1 year group. Distension of the biceps brachii tendon sheath occurred in 8/32 (25.0%) patients with arm swelling within 1 year and 6/15 (40%) patients with arm swelling after 1 year. Adhesive capsulitis arose in 7/32 (21.9%) of patients in the within 1 year group and 4/15 (26.7%) patients in the

Table 2 - Clinical features of the shoulder disease in the study subjects.

	Number of patients of clinical feature	
	Arm swelling duration ≤ 1 year (n=32)	Arm swelling duration > 1 year (n=15)
Abnormalities of ultrasound findings	27 (84.4%)	14 (93.3%)
SST tear	5 (15.6%)	8* (53.3%)
SASD bursa thickening	20 (62.5%)	6 (40.0%)
Distension of BB tendon sheath	8 (25.0%)	6 (40.0%)
Adhesive capsulitis	7 (21.9%)	4 (26.7%)

SST, supraspinatus tendon; SASD, subacromial subdeltoid; BB, biceps brachii. *$P < 0.05$.

after 1 year group. Patients with a supraspinatus tendon tear (n=13) revealed a significant longer duration (1310 days vs. 398 days, $P=0.026$). Subacromial subdeltoid bursal thickening had shorter duration trend but was not significant (454 days vs. 892 days, $P=0.058$). (Fig. 1)

DISCUSSION

Our current study findings demonstrate that the pathology of shoulder pain in patients with BCRL is related to the duration of the lymphedema. We found from our analysis that the lymphedema duration influences the rotator cuff tendon pathology, but that the symptoms do not correlate. It will therefore be important to further examine any correlation between radiological findings and the clinical features of shoulder disorders among breast cancer patients after therapy.

Several studies have reported various incidences of shoulder pain (from 15% to 28%) in patients surgically treated for breast cancer [10-13], but few of these reports have assessed patients with BCRL. The incidence of shoulder pain in BCRL cases may be increased because it may cause or aggravate this complication. In our present study, we observed that 53% of our patients with BCRL had ipsilateral shoulder pain, similar to that of a previous study that reported an incidence of 71% [5]. The incidence of shoulder pain was higher (59.3% vs. 40%), and the visual analog scale was significantly higher in the within 1 year group of patients in our current study. We believe that the early period of lymphedema may be at high risk for pain sense because the patient would have undergone adjuvant chemo- and/or radio-therapy during this time. Chemo-therapy induced fatigue, bone pain and/or shoulder position for radio-therapy could also affect shoulder pain. Moreover, in the early period of lymphedema, the patients may be more sensitive to pain due to inflammatory processes and this could affect other shoulder pathologies.

Ultrasound is a useful examination methodology for patients with BCRL. Skin thickness is measured using ultrasound in our clinic, as are the shoulder rotator cuff regions so that appropriate exercise programs can be prescribed. Shoulder exercise programs are one component of complex decongestive physiotherapy [14]. Herrera and Stubblefiedl [2] have reported that rotator cuff tendinopathy is a complication of lymphedema caused by an internal disruption to the arrangement of tendon fibers. Mellor et al. [15] have documented that the measurement of skin thickness using ultrasound may be a useful clinical tool for the diagnosis of lymphedema and may also assist with further investigations of therapeutic techniques.

BCRL patients commonly present with shoulder discomfort and a previous study [5] has reported that 21.1% of the BCRL cases analyzed had a supraspinatus tendon tear. We found in our present analyses that 27.7% of our BCRL cases had a supraspinatus tendon tear, which was more common in patients who developed arm swelling after more than 1 year and was found to be significantly related with arm swelling duration. We believe that a lymphedema can impact on the rotator cuff pathology through chronic inflammation and immobilization. In addition, a longer lymphedema duration may have a cumulative effect on the rotator cuff pathology through a traction effect, thus providing a possible explanation as to why a supraspinatus tendon tear was more common in the after 1 year group. In the present study, there was much more patients with SASD bursal thickening compared to a previous study (55.3% vs 5%) [5]. This disagreement may be caused by the different ultrasound diagnosis criteria. We included the SASD bursal thickening as the pathologic findings of ultrasound, however Jeong et al. [5] used subacromial bursitis as the pathologic findings. It may make a discrepancy of overall abnormal ultrasound rate between two studies as well.

On the other hand, some of our BCRL patients had complained of contra-lateral shoulder pain also, which may be caused by an overuse of the contra-lateral arm due to a non-use of the BCRL involved arm. As this contra-lateral shoulder could show different pathologic features to the BCRL involved arm, further radiologic studies of contra-lateral shoulder pain are needed in BCRL patients.

Adhesive capsulitis is also a common finding in BCRL patients. A previous study [5] has reported that 21.1% of the BCRL patients analyzed had adhesive capsulitis. Adhesive capsulitis is characterized by pain and a loss of stiffness in the shoulder, and its prevalence is about 2% in the general population [16]. We found in our current BCRL cohort that 23.4% of the patients had adhesive capsulitis, but that the duration of lymphedema was not significantly different in these cases.

There was no significant correlation found in our analysis between the results of the DASH questionnaires and the duration of lymphedema. However, our small sample size could have limited our ability to undertake a robust assessment of this relationship and further studies are needed in this regard. We recognize also that there are some other limitations of our present study. First, we assessed a single center cohort with, as mentioned, a relatively small number of patients. Second, because almost of patients were International Society of Lymphology lymphoedema stage II and just two of 13 patients with a supraspinatus tendon tear had a full thickness tear, we could not investigate the relationship between the severity of lymphedema and the severity of shoulder pathology. Further study would be expected. Third, the age and type of surgery between the two groups we analyzed were different. We could not rule out that a degenerative rotator cuff pathology would have occurred in some patients regardless of BCRL.

In conclusion, our current findings suggest that patients who experience an early period of BCRL more commonly show ipsilateral shoulder pain and that BCRL can have a cumulative impact on the rotator cuff pathology. We thus recommend that clinicians should adopt an early management approach to shoulder pain in BCRL cases by making a precise diagnosis of this complication.

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STEWART-TREVES SYNDROME: CASE REPORT

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Competing interests
The author declare no competing interests.

ABSTRACT

F. Stewart and N. Treves in 1948 first describe extremely aggressive lymphangiosarcoma with a poor prognosis that develops due to radical mastectomy and radiotherapy in post longtime - lymphedema. Currently, approximately 400 cases of Stewart-Treves syndrome are reported in world literature. Prevalence is 0.45% in patients who survive longer than five years after a mastectomy to breast cancer. Diagnosis is based on biopsy of the lesion and immunohistochemical analysis. Sentinel lymph node biopsy, prevention and follow up of post mastectomy lymphedema reduce incidence of Stewart-Treves syndrome. We describe the case of a woman who developed angiosarcoma in post longtime - lymphedema after mastectomy with adjuvant radiotherapy (10 years).

Keyword

Stewart-Treves syndrome - Postmastectomy angiosarcoma - Lymphangiosarcoma - Breast cancer - Lymphoedema

CASE HISTORY

G.F. 67 year old woman, 3 pregnancies to term, anamesis negative family to breast cancer. In March 1995 modified radical mastectomy dx, axillary lymph node clearance and multiple cycles of chemioterapy for invasive ductal carcinoma average degree of differentiation with axillary lymph node metastases (pT2, pN1; M0). Patient developed clinically important lymphedema in addition to repeated lymphangitis and erysipelas whit lymphorrhoea. In May 2005 has come to our Clinic for cronic post longtime lymphedema (10 years) right arm stage III painful and functional limitation. The arm volume had increased 68.3% compared to the contralateral (Fig. 1-2-3). Patient presented fibrosis, skin tense, nodular purplish and yellow-brown crust lesion wrinkled appearance, small hemorrhagic areas and marginal telangectasica area (Fig. 4-5) neck and chest unscathed. Antibiotics, anti-inflammatory phlebotonics drugs and decongestiva combined physiotherapy decreased minimally arm volume but not substantially improved the symptoms. Patient was submitted to biopsy, immunohistochemical diagnosis was angiosarcoma: sections of skin with irregularly shaped vascular cavity in the dermis and subcutaneous bounded by atypical cells of epithelioid forms that express the factor VIII and CD31, negative for cytokeratin and S-100 protein. Amputation of the affected limb was discussed but she did not wish to pursue this option and was placed on palliative pain control. The exitus occurred about 16 months later. (September 2006).

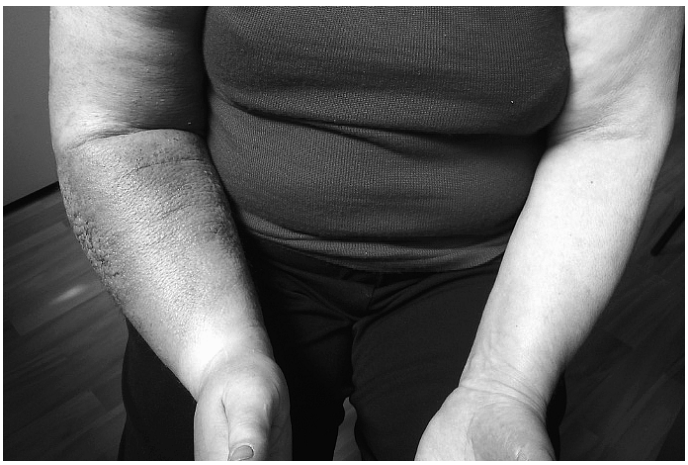


Figure 1

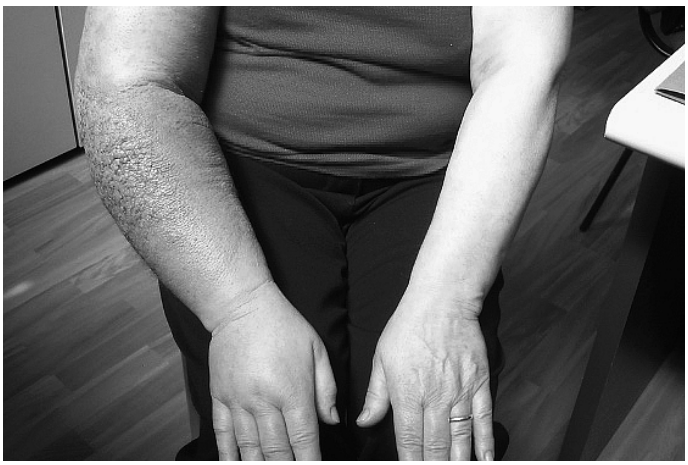


Figure 2

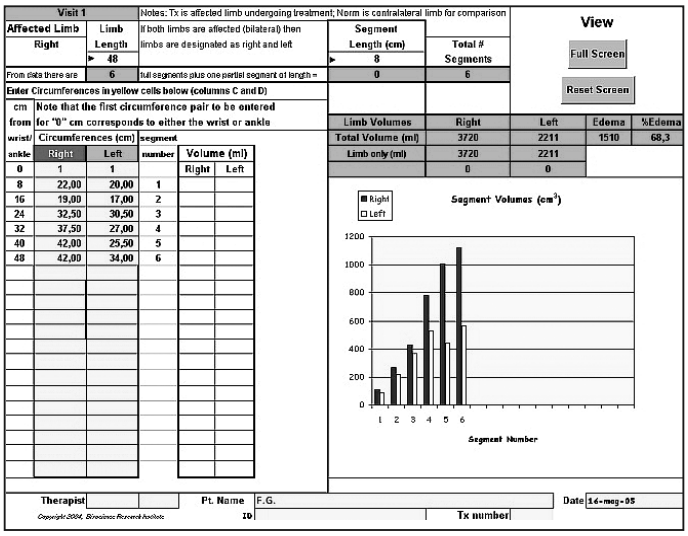
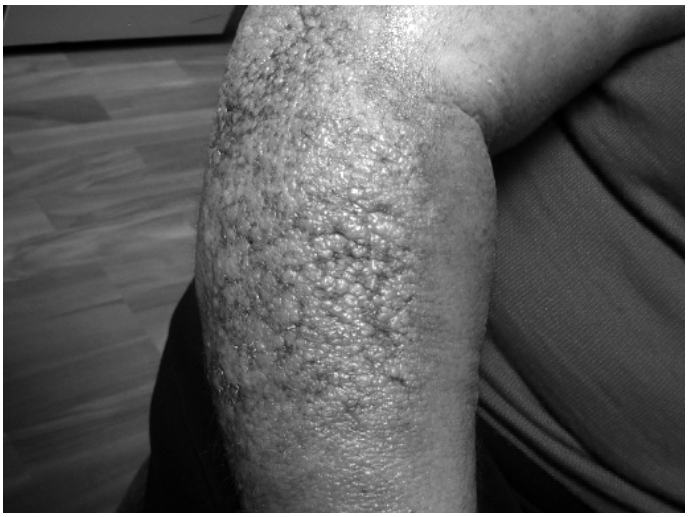


Figure 3



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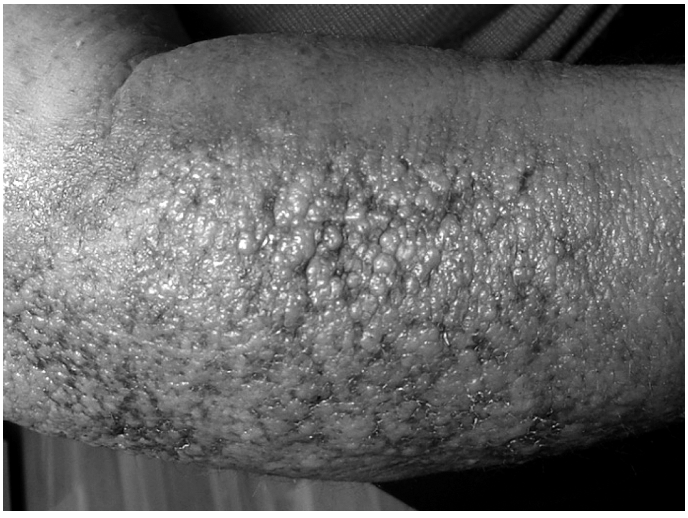

Figure 5

DISCUSSION

Stewart-Treves sindrome is lymphangiosarcoma of high-degree malignancy and poor prognosis. It manifest as telangiectasia or ecchimosis with rapid growth, which could coalesce and produce plates or cutaneous nodules. It can present ulceration and cause pain depending on the relationship with the vascular-nerve bundle of the arm [4]. Present local diffusion through subcutaneous for the adjacent structures and early systemic hematogenic diffusion mainly for the lung [3]. The most common location is the arm, followed by the forearm, elbow and thoracic wall [3]. The average time interval between mastectomy and lymphangiosarcoma development is 10,3 years in accord to our case report. Etiopathogenesis is related to chronic lymphedema generates hyperstimulation to the formation of new lymphatic vessels and predisposes the emergence of cellular mutations of the endothelial of vessels leading to the development of angiosarcoma [2].
The standard treatment consists of resection of the lesion with three-dimensional wide to limb preservation [3]. In the cases that resection with preservation of the limb is not possible (invasion of the vascular-nerve bundle) the surgery consists of amputation.

CONCLUSION

Stewart-Treves syndrome is a rare aggressive lymphangiosarcoma with a poor prognosis that develops in post longtime - lymphedema arm after mastectomy (> 5 years) Early diagnosis and promptly association with surgical therapy improves prognosis.
Sentinel lymph node biopsy, prevenction, decongestive combinate physiotherapy, elastic compression sleeve and follow up of post mastectomia lymphedema decrease incidence of Stewart-Treves syndrome.

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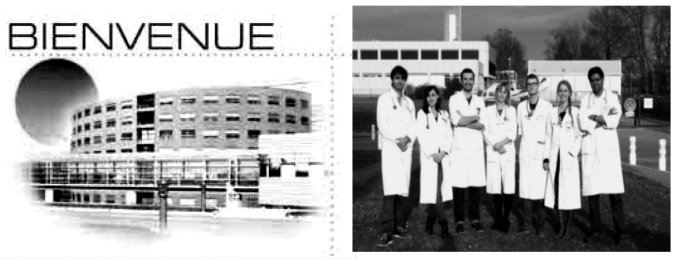

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With warm regards,

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