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A RETROSPECTIVE STUDY OF THE EFFECTS OF THE LYMPHAPRESS PUMP ON LYMPHEDEMA IN A PEDIATRIC POPULATION

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ABSTRACT

We studied the effects of the Lymphapress pump (LP; Global Medical Imports, Digby, NS, Canada) retrospectively on 16 children with primary or secondary lymphedema of the upper or lower extremities by measuring the volume and circumference of the limbs before and after treatment. We reviewed medical charts for data on age, sex, length of disease process, grade of lymphedema, frequency and duration of treatment, and pump pressures used.

We recorded changes in limb size before and after pumping in terms of the mean percentage difference between the affected and unaffected limb at both time points to allow for growth of the child and the extremity.

On volumetric measures, thirteen (93%) of the subjects showed a clinical trend towards sustained maintenance or reduction in size of the lymphedematous limb(s).The reduction in the pump pressure at start of the treatment to that required to maintain the size of the limb was statistically significant ($p = 0.0036$). Fourteen (88%) of the subjects had no complications directly attributable to the pump, whereas two had complications that were probably unrelated to LP. Overall, there was a clinical trend towards reduction or maintenance of the lymphedematous limb size in children using LP without notable adverse sequelae.

Toronto's Hospital for Sick Children (HSC) manages a large population of children including those with lymphedema, a comparatively rare but troublesome condition. Physiotherapy tries to control edema in limbs to restore function and prevent chronic skin changes. Two of the more common management techniques are multi-sequential external pneumatic pumps, such as the Lymphapress pump (LP, Global Medical Imports, Digby, NS, Canada) and a manual massage regimen generally termed complex physical therapy (CPT). Clinicians at HSC who have used the LP describe excellent outcomes with no notable side effects. Advocates of CPT (1,2), on the other hand, maintain that use of LP leads to development of fibrous bands which further impede lymph drainage or produce lymphedema in the unaffected limb. However, the effectiveness of physiotherapy in the management of lymphedema has not been documented. No randomized controlled trials in adults or children have been reported. No study of these two therapies has produced statistically significant findings in favor of one over the other.

The sequential compression action of the LP drains lymph from the affected area to the more proximal lymphotome (3). Proponents of CPT claim that normally functioning lymphatics need to be "activated" before treatment of the affected limb, first in the

contralateral quadrant of the trunk and then in the ipsilateral (2,4). This activation ostensibly promotes drainage of lymph through dilated lymph vessels across the lymphatic watersheds (2). Without such drainage, excess fluid can be displaced into the adjacent lymphotome, thereby increasing localized edema (1,4). Some researchers suggest that pumping increases edema by forcing water out of the affected area, leaving behind concentrated tissue proteins that generate chronic inflammation and fibrosclerosis, with more edema (1,2,4-7). The effectiveness of pumping is also hindered by the degree of subcutaneous fibrosis. These major changes mean that grade III edemas, in particular, can be resistant to treatment (8,9). One study that monitored patients for 2 years reported that use of pumps decreased the circumference of the affected limb without complications (8). The same study noted clinically significant decreases in limb circumference at the calf level and below.

The optimal pump pressures and compression cycle, however, continue to be debated (3,9-12). One study suggests that the initial lymphatics and superficial collecting lymphatics are damaged by exposure to pressures of 60 to 70 mmHg and above, which leads to fibrosis and further lymphatic blockage (l).

The effectiveness and possible complications of LP use need to be clarified, particularly in treatment of children. The purpose of this study was twofold: to determine (1) whether there was a sustained reduction and/or maintenance in limb volume and circumference with the use of a multisequential compression pump in our pediatric population, and (2) whether the complications attributed to these pumps occurred in our subjects.

MATERIALS AND METHODS

We drew our sample from a population of children with a diagnosis of primary or secondary lymphedema treated at HSC between 1987 and 1997. We retrospectively reviewed the medical and physiotherapy charts of 16 boys and girls (7 and 9, respectively) with lymphedema, either unilateral or bilateral. Because of the comparative rarity of the disease in children, some patients were followed into adulthood before transferring from HSC to a general hospital. Patients with mixed conditions, such as Klippel-Trenaunay-Weber syndrome, were excluded because of compounding features such as blood vascular abnormalities. The publication of this paper was approved by the Hospital's Research Ethics Board.

The principal investigator reviewed the medical charts for data on age, gender, grade of lymphedema according to Földi et al (4), age of diagnosis, and age at which treatment with LP began. The LP regimen devised by a physiotherapist at HSC consisted of pumping, wearing of graded compression garments, and advice on exercise and skin hygiene. Frequency and duration of treatment, pressures used, and number of times per week spent pumping to maintain the size of the limb were also noted.

Measuring changes in limb size due to therapeutic intervention is more difficult in children, as normal growth and development take place at the same time. Few studies have addressed this issue, and in the absence of a reliable, generally accepted standard, HSC clinicians have developed their own system to measure these changes. It entails measuring the volume (ml) of water displacement at either the groin (most medial, superior aspect) or the axillary level of the affected and unaffected limb. The percentage difference between measurements of volume is calculated by subtracting the result at completion of the regimen from that at the start. The staff physician and physiotherapists who treated the patients during the study considered a 10% difference to be clinically significant. A difference greater than 10% indicated improvement: i.e., the lymphedematous limb decreased in size compared with the unaffected one. Less than

10% difference showed maintenance, and a difference of more than -10% indicated deterioration, that is, the affected limb increased in size compared to the unaffected limb.

Using a tape, we measured circumference (cm) at 7 points on the lower extremity (LE) (groin, thigh, calf, 3 cm above ankle, ankle joint line, midfoot and toes, using the patellar base as a reference point) and 8 points on the upper extremity (UE) (apex of axillary fold, mid-upper arm, lower upper arm, elbow crease, mid-forearm, lower forearm, wrist crease and hand, using the elbow and wrist creases as reference points). From these data we estimated a mean total limb circumference. The percentage difference in the mean circumference of the affected and unaffected limbs was calculated before the start of the LP regimen and on the last recorded visit to HSC. The clinicians treating the patients considered a 5% difference in circumferential measurements before and after pumping to be clinically significant. A difference of less than 5% indicated that the size of the affected limb was maintained. A difference greater than 5% indicated that the affected limb decreased in size compared with the unaffected limb. A deterioration was implied if there was a difference of more than -5%. The differences in measurements of volume and circumference in a single LE lymphedema were plotted. Measuring the treatment effects on bilateral lymphedemas is difficult, as there is no control limb for comparison. In the absence of a reliable standard, we devised a method of measuring the change in volume by comparing thigh-to-foot ratios in the same limb. Right and left extremities could then be compared before and after LP. We estimated that a difference of more than 2 in the ratio before and after pump use showed improvement, a decrease of more than 2 showed deterioration, and a value in between showed maintenance.

Complications, noted as present or absent, were the development of fibrosclerotic bands, skin lesions varying from the development of small, red, warm areas to open sores, lymphedema of the unaffected limb if unilateral, genital or truncal lymphedema, and throbbing or painful sensations in the lymphedematous limb. Complications present before the pump treatment started were not included.

Subject data and the LP regimen characteristics were analyzed for means and standard deviations. Paired Student's t-tests were used to analyze pump data on volume, circumference, and pump pressure before and after treatment. A p value of 0.05 was considered significant. Data were also analyzed for clinical trends, based on the experience of the expert clinicians involved in the treatment of this disease.

RESULTS

Table 1 summarizes the data on subject characteristics and LP treatment. The mean age at diagnosis was 5.2 ± 5.06 years (mean ± standard deviation; range, 0.1 to 14 years). Pump pressures varied from 50 to 140 mmHg at the start of treatment regimen with a mean of 84.29 ± 24.72 mmHg. During the maintenance phase of treatment, pressures were reduced to between 35 and 100 mmHg, with a mean of 58.57 ± 19.05 mmHg. The difference between pump pressures before and after treatment was found to be statistically significant ($p = 0.0036$) *(Fig. 1)*. Pump volume and circumference data in both the UE and LE groups before and after the regimen did not differ statistically. However, several clinical trends were identified. Our measurements of volume in 15 of the 16 subjects (one subject was unable to comply with volumetric measurements) showed that 20% of affected limbs improved, 73% were maintained, and 7% deteriorated. No complications directly attributable to the pump were found in 88% of subjects.

Measurements of Volume and Circumference in the Lower Extremities

TABLE 1
Subject Characteristics and Results of LP Treatment

Variables	All Subjects n = 16	Unilateral UE n = 3	Unilateral LE n = 7	Bilateral LE n = 6
Sex Male	7	2	2	3
Female	9	1	5	3
Age (yrs)				
Mean (SD)	13.94 (5.14)	9.33 (2.08)	16.71 (5.38)	13.00 (4.29)
Range	6 - 26	7 - 11	9 - 26	6 - 18
Diagnosis age (yrs)				
Mean (SD)	5.20 (5.06)	1.13 (0.75)	6.43 (5.74)	6 (4.72)
Range	0.10 - 14	0.60 - 2	0.10 - 14	0.30 - 11
LP start age (yrs)				
Mean (SD)	7.93 (5.48)	2.67 (2.08)	10.43 (5.65)	7.85 (4.55)
Range	0.80 - 16	1 - 5	0.80 - 16	1.10 - 13
Initial LP pressure				
Mean (SD)	84.29 (24.72)	70.00 (0.00)	88.60 (30.8)	81.67 (19.41)
Range	50 - 140	70 - 70	50 - 140	50 - 100
Pressure reduction				
Mean (SD)	58.57 (19.85)	75.00 (21.20)	63.57 (21.74)	45.00 (7.07)
Range	35 - 100	60 - 90	40 - 100	35 - 50
Treatment duration (months)				
Mean (SD)	55.81 (22.01)	68.30 (25.40)	50.14 (18.67)	54.00 (23.04)
Range	19 - 96	46 - 96	23 - 75	19 - 84
Times per week				
Mean (SD)	6.68 (0.70)	6.33 (1.15)	6.85 (0.37)	6.66 (0.81)
Range	5 - 7	5 - 7	6 - 7	5 - 7
Maintenance times per week				
Mean (SD)	5.33 (1.95)	7.00 (0.00)	4.42 (2.22)	6.00 (1.67)
Range	2 - 7	7 - 7	2 - 8	3 - 7
Hours per day				
Mean (SD)	3.96 (2.09)	4.67 (2.08)	3.30 (2.33)	4.16 (2.13)
Range	1 - 8	3 - 7	1 - 7	2 - 8

LP=Lymphapress pump; SD=standard deviation; UE=upper extremity; LE=lower extremity; yrs=years

The volume of the unilateral LE lymphedema in the affected limb of subject 2 was greater than in the unaffected limb, indicating a deterioration *(Fig 2)*; the volume of the unilateral LE lymphedema in the affected limb of subject 4 was less than in the unaffected limb, indicating an improvement; and the volume of the affected limbs of subjects 1,5,6, and 7 showed less than a 10% difference between their respective unaffected limbs, indicating maintenance.

The circumference measurements of subjects with unilateral LE lymphedema (subjects 1,2, 5, and 6) showed less than a 5% difference between the affected and unaffected limbs *(Fig. 3)*, indicating maintenance. The

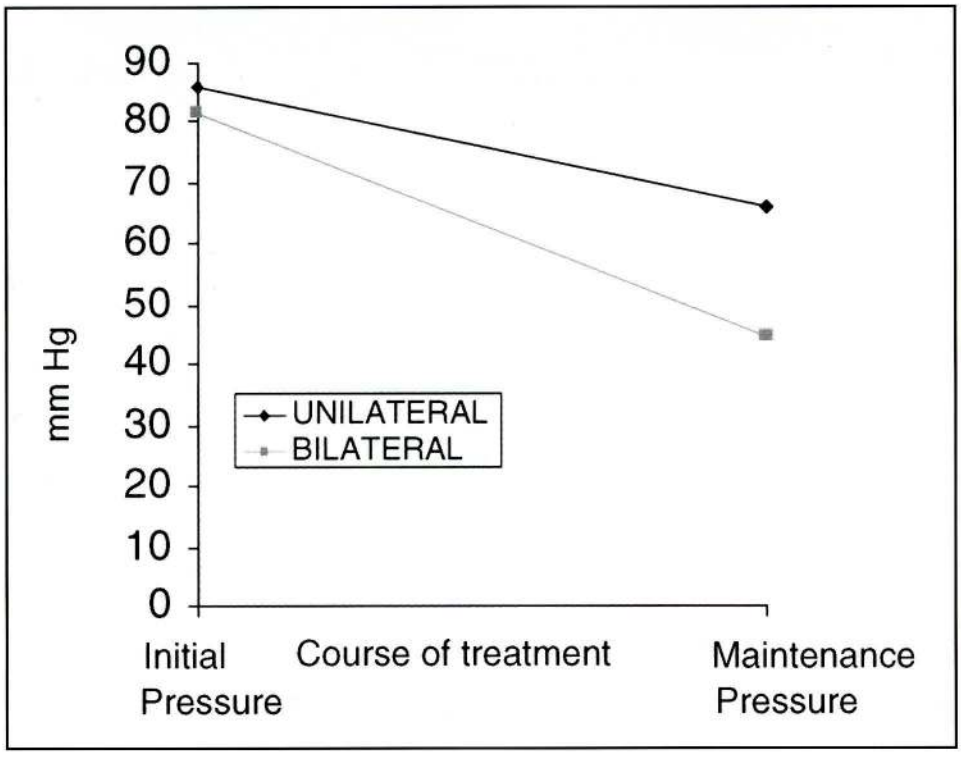

Fig. 1. Pressure reductions expressed as a mean from start of treatment to level needed to maintain limb size in subjects with unilateral and bilateral lymphedemas (p = 0.0036, by paired Student's t-test).

affected limbs of subjects 3,4, and 7 decreased in size compared to the unaffected limbs, indicating improvement *(Fig. 3)*. Over a period of 23 months, the affected limb of subject 6 became smaller than the unaffected one, but still remained within the maintenance range.

Measurements of Volume and Circumference in the Upper Extremities

The volume measurement of the unilateral UE lymphedema of subject 1 increased by 28%, indicating that the affected limb increased in size over the unaffected limb. There was less than a 10% difference between the volumes of the affected limb and unaffected limb of subject 2, indicating maintenance. The size of the affected arm of subject 3 decreased in size by 16.3%, indicating improvement.

Measurement of circumference of unilateral UE in subjects 1 and 3 showed maintenance, whereas subject 2 showed deterioration with a negative value of -7.24%.

Bilateral LE Lymphedema

In all 6 subjects with bilateral LE lymphedema, the ratios between right and left limbs showed maintenance, as there was no increase or decrease greater than 2 *(Fig. 4)*.

Complications

Two patients or 12% had complications. One boy with LE lymphedema moved to a warmer climate and developed an infection in the affected limb. A girl with LE lymphedema, and comorbid conditions of cerebral palsy and microcephaly, developed a pressure sore on the heel of the lymphedematous foot.

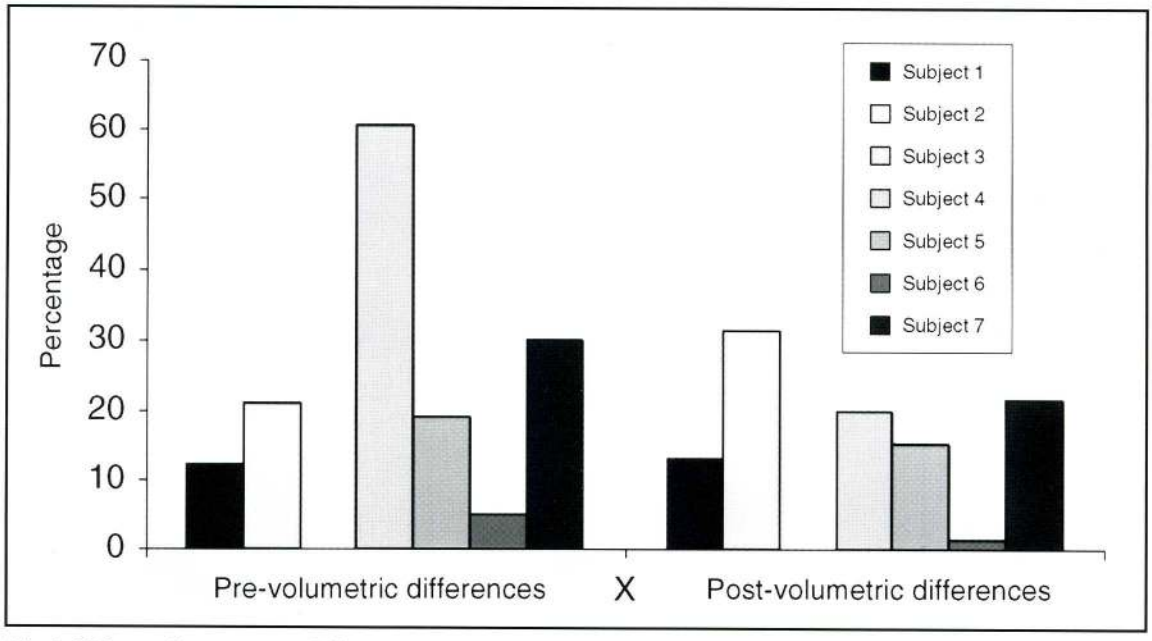

Fig. 2. Volumetric percentage differences between the lymphedematous lower extremity and the normal limb before and after treatment with the Lymphapress pump in seven subjects with unilateral edema.

Two subjects had pre-existing complications. One boy with bilateral LE lymphedema had scrotal edema before starting LP treatment. Another boy with edema of the face, chest, scrotum, and abdomen had these swellings from birth. Pumping was carried out from the age of 2 to 6 years, then stopped because of problems with compliance. These pre-existing complications were excluded from the data analysis because they were not related to pump usage.

DISCUSSION

This study was limited by the small number of subjects and the lack of rigorously standardized measurement tools. However, the findings provide useful information about the effectiveness of the LP and the potential complications attributable to its use for clinicians managing this troublesome disorder in children. Certain clinically significant trends noted in this study are discussed below.

Measurement of volume change is a valid method of quantifying altered limb size and more reliable than measurement of circumference (13-17).

One girl with a UE lymphedema showed a -28% difference in pump volumetric measurements, indicating an increase in size of the affected arm. It is speculated that noncompliance with wearing a compression garment that formed part of the treatment reduced its effectiveness. Another girl (subject 6) with unilateral LE lymphedema showed a decrease in circumference of the affected leg compared with the unaffected leg of -1.45%, a decrease in size, perhaps due to muscle wasting. Another explanation, suggested by Casley-Smith et al (18), is that the "unaffected" limb is also slightly lymphedematous, but the edema is subclinical. The known affected limb may therefore appear to become smaller than the other. However, in our study, volumetric measurements of the same subject showed a decrease of 3% in size of the affected limb, indicating maintenance and

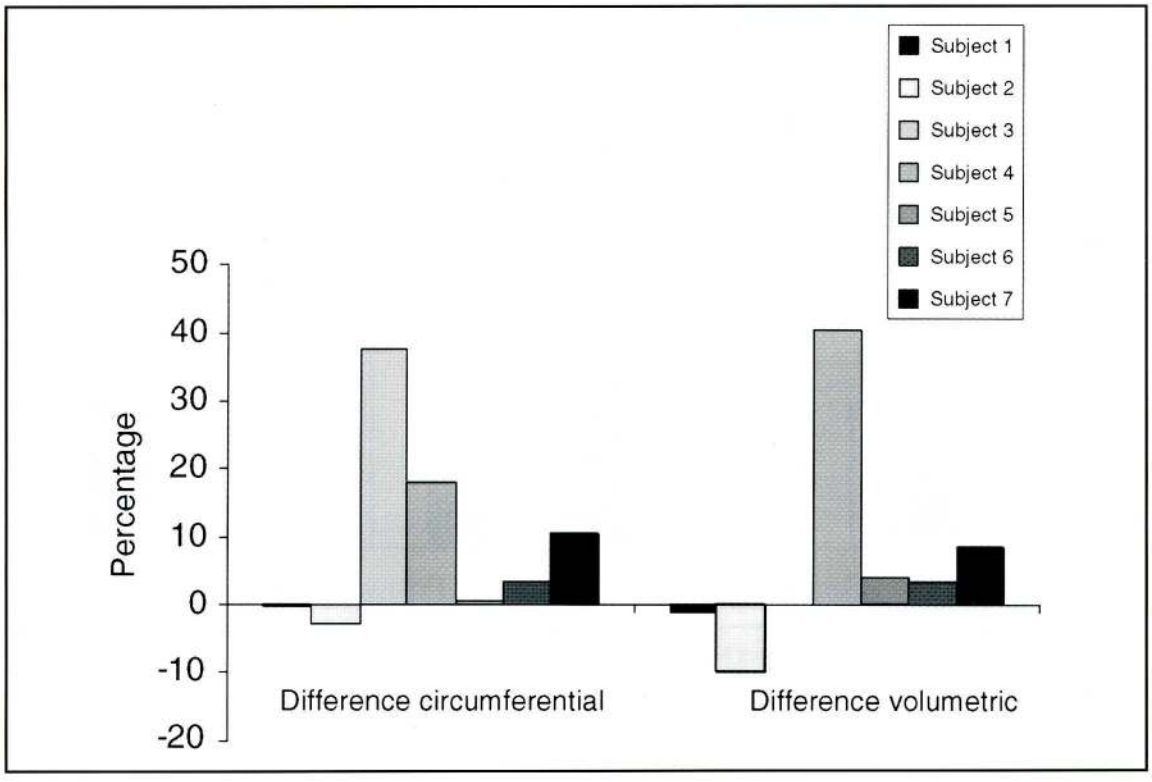

Fig. 3. Changes in circumferential and volumetric measurements in the seven subjects with unilateral edema of the lower extremity (LE) after use of the Lymphapress pump, expressed as a percentage difference between the lymphedematous LB and the normal limb. A negative value indicates an increase in size of the affected limb.

therefore a discrepancy between the two methods of measurement *(Fig. 3)*.

Most studies of the reliability and validity of measures of volume and circumference have examined adults, whose limb size remains constant, whereas the growing child presents more challenges. One study (13) compared the reliability and validity of a computerized limb measurement system (CLEMS) with other methods of measuring lower limb volume. It reported a strong correlation r = 0.992) between CLEMS and the water displacement method of measuring limb volume. Both methods, however, showed a poor correlation with the tape measure method r = 0.318 for water displacement, 0.341 for CLEMS). The study concluded that the tape measure method was inaccurate for measuring lymphedema volume. Another study (14) reported discrepancies between volumes calculated by water displacement and those based on measurements of circumference. In the LE, volumes calculated from measurements of circumference with the truncated cone method showed only half the amount of edema compared with the water displacement method. In the UE, the two methods produced near identical values. This study hypothesized that this result occurred because the UE tends to swell uniformly and the LE swells disproportionately more distally. Therefore, when the LE is measured, a certain amount of normal tissue is included in the measurement. Stranden (17) asserts that volumes calculated from surface measurements in adults are accurate only for clinical use. Measurements of circumference may help identify specific problem areas on

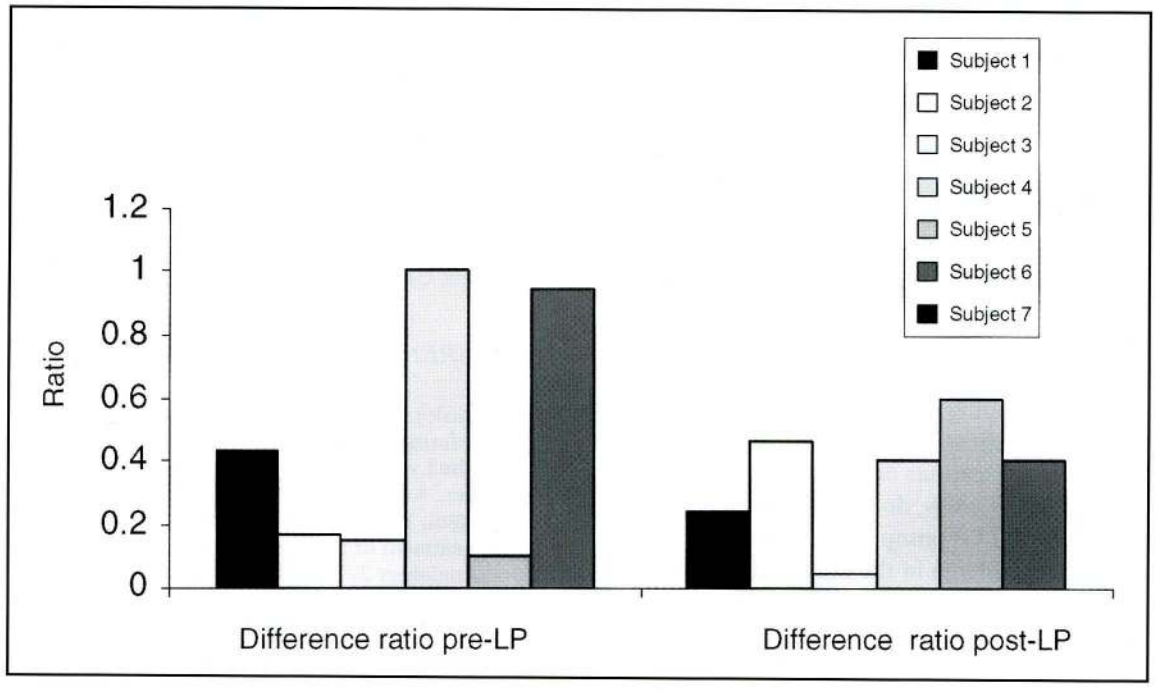

Fig. 4. Differences in thigh-foot ratios between right and left lymphedematous extremities before and after treatment with the Lymphapress pump (LP), in six subjects with bilateral edema. Ratios were calculated from measurements taken at the groin and ankle.

the lymphedematous limb and the response to treatment in these areas compared with the other limb, or to identify where measurements of volume are not possible (14). In short, we have found that volumetric measurements are suitable for measuring changes in limb size in children, whereas superficial circumferential measures and other similar formulas fail to take into account normal limb growth.

No studies have examined interrater consistency in taking volumetric measurements, although repetition in measuring improves reliability (19). Three of the four therapists in our study were experienced at taking volumetric measurements but interrater reliability among them was not tested, and therefore could have influenced the results.

In our study, measures of volume indicated that use of the LP tends to decrease or maintain the size of the lymphedematous limb in a pediatric population. Differences of 10% in volume measurement and 5% in the mean circumference and the thigh-to-foot ratios in both limbs were used to indicate improvement, maintenance, or deterioration in the absence of an accepted standard. These percentages were arbitrarily determined by an expert physical therapist and physician in the field of childhood lymphedema to account for differences in limb size in the growing child. Clinical experience cannot be underestimated because, in the absence of more sensitive measures, it is used to make decisions about the LP regimen, such as decreasing the pumping pressure.

Our study found a significant difference between the initial pump pressure used to reduce edema and the amount needed for maintenance. Regarding pump pressures, Casley-Smith et al (1) claimed that superficial capillary plexuses were broken at pressures greater than 60 mmHg, leading to fibrosis

and progressive lymphatic blockage. Although the total mean pumping pressures decreased from 84.29 to 58.57 mmHg, it was not possible to show from this study whether lymphatic structures were, in fact, damaged. Subsequent to this study, the pressures used were reduced to between 40 to 60 mmHg to accommodate tolerance levels and decrease the risk of damage to lymphatic structures.

Most (88%) of our subjects experienced no complications directly arising from pump use. However, it is impossible to demonstrate whether pumping or some other factor caused the two complications identified. The more serious complications of lymphedema, genital and truncal edemas, do not appear to be caused by the LP pump itself, nor does it cause lymphedema in the unaffected limb. These conditions existed in two subjects before pump treatment commenced. However, it is not known whether the pump exacerbated the genital edema. The boy with bilateral LE lymphedema and edema of the groin and scrotum was reluctant to discontinue pumping despite these regional swellings, because he had a decrease in infectious episodes while on treatment. It remains unclear whether pumping when these comorbid conditions are present is harmful.

CONCLUSION

The Lymphapress pump is useful to maintain or reduce the size of a lymphedematous limb in a child without notable complications. We also were able to substantially reduce the amount of pump pressure required to treat the lymphedematous limb. However, more research is needed at a microcirculatory level on the effects of compression on lymphatic structure in both children and adults. The validity and reliability of the thigh-to-foot ratio measures used to evaluate bilateral lymphedema also needs to be further evaluated. Finally, further research is required to identify whether the Lymphapress pump, complex physical therapy, or a combination of the two regimens is the optimal non-operative method to control childhood lymphedemas.

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