

## The Godoy & Godoy cervical stimulation technique in the treatment of primary congenital lymphedema

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### Abstract

The aim of the current study is to report on the treatment of primary lymphedema using a new form of therapy: cervical stimulation. In a prospective cohort study, 9 boys and 5 girls with primary congenital lymphedema were evaluated over two years. Age ranged from two months to 8.5 years. After diagnosis, all mothers were trained in the new technique. The Godoy & Godoy cervical stimulation technique consists of between 20 to 30 stimuli per minute using light movements in the cervical region. All the children were submitted to perimetric evaluations of the feet and legs; however, in this study only two points, 3 and 6 cm from the base of the big toe nail, were used. The two-tailed t-test was used for statistical analysis with an *alpha* error of 5% ( $P < 0.05$ ) considered acceptable. The size of the lymphedematous feet was reduced and even normalized ( $P < 0.0001$ ) for all treated children. Cervical stimulation is a new option in the treatment of primary congenital lymphedema; its association with compression stockings has a synergistic effect in reducing the volume of lymphedema.

### Introduction

Lymphedema, defined as the abnormal accumulation of protein-rich fluid in soft tissues, is a result of dysfunction of the lymphatic system that causes an imbalance between lymph formation and its absorption into the initial lymphatics.<sup>1,2</sup> Another definition is a progressive, but usually painless, swelling of the limbs or genitals that is the result of a decrease in transport capacity of the lymphatic system.<sup>3</sup> Lymphedema in children is rare with a reported prevalence of approximately 1.15:100,000 in those under 20 years of age.<sup>4</sup> Another study of 312 index patients with pri-

mary lymphedema under the age of 36 years reported a frequency of 1:6000 at birth in a ratio of one male to three females.<sup>5</sup>

Intra-familial clinical heterogeneity has been reported, as has variability in age at onset. There are more than four known genetically distinct lymphedema conditions and mutations of three genes have been discovered in families with congenital lymphedema.<sup>6</sup> Lymphedema of the lower extremities is a diagnostic challenge with the first step being the exclusion of secondary causes of swelling and secondary lymphedema. The majority of cases of primary lymphedema are due to inborn abnormalities of the lymphatic system.<sup>3</sup> Primary lymphedema is classified as sporadic/non-familial *versus* familial lymphedema in one group of congenital defects. Findings support the recommendation that a further careful and comprehensive clinical evaluation is warranted, including a detailed investigation of dysmorphic features together with imaging of the lymphatic system with a subsequent syndromic classification when peripheral lymphedema of undetermined etiology is found.<sup>7</sup> In long-lasting lymphedema, interstitial accumulation of proteins, inflammatory cells, adipose tissue hypertrophy and fibrosis make pitting less evident.<sup>8</sup> In primary congenital lymphedema, treatment focuses on non-surgical options, such as manual lymph drainage, compression therapy and exercises closely involving parents. However, few studies report the evolution in the role of patients with respect to the different forms of treatment given. The aim of the current study is to report on the results of the treatment of primary congenital lymphedema over a period of two years using a new form of treatment.

### Case Report

Fourteen children (9 boys and 5 girls) with primary congenital lymphedema were evaluated in a prospective cohort study carried out from 2004 to 2009 in the Godoy Clinic, Brazil. Age at start of treatment ranged from two months to 8.5 years. Inclusion criterion was the presence of clinically diagnosed lymphedema of a lower extremity confirmed by perimetric measurements at two points, 3 and 6 cm from the base of the big toe along the dorsum of the feet. Four children were excluded from the study because they were unable to periodically return for treatment due to the distance from their homes to the treatment center. After diagnosis, all the mothers were trained in the manual cervical stimulation technique and,<sup>9-11</sup> when considered sufficiently proficient, they were given permission by the therapeutic team to continue treatment in their own homes. Mothers

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were requested to perform stimulation for 15 to 20 min per day at 20 to 30 stimuli per minute using light movements.<sup>9-11</sup> For some children, a home-made compression stocking made of a cotton-polyester material was used. Adjustments were made to the size of the compression stocking as the volume of edema was reduced.<sup>11</sup> All the children were submitted to perimetric evaluations of the feet and legs, although for this study, only measurements of the feet at two points, 3 and 6 cm from the base of the big toe along the dorsum of the feet, were used. The edema was evaluated at all clinic visits. Mothers were encouraged to take part in these evaluation sessions so that they participated more in the treatment of their child. Low-stretch cotton-polyester compression stockings were used in the treatment of 4 children. The two-tailed t-test and Wilcoxon's matched pair-test were used for statistical analysis with an *alpha* error of 5% ( $P < 0.05$ ) considered acceptable. The study was approved by the Research Ethics Committee of the School of Medicine in São José do Rio Preto (FAMERP protocol n. 194/2009). After the nature of the study had been explained, the parents or guardians of the children signed informed written consent forms.

### Results

At the start of treatment, perimetry revealed a statistically significant difference between the feet affected by lymphedema and those not affected in 10 children (paired t-test  $P < 0.0001$ ) (Table 1). After two years of treatment there was less of a difference, but this

was still significant (paired t-test  $P < 0.02$ ) (Table 2).

A significant reduction in the perimeter of feet was identified on comparing the size of the foot before treatment and after two years of treatment using only cervical stimulation (paired t-test  $P = 0.0001$ ) (Table 3).

For 4 children who underwent the combined treatment (cervical stimulation and compression garment), perimetry revealed a significant difference on comparing the foot affected by lymphedema with the unaffected foot before treatment (paired t-test  $P < 0.0002$ ) (Table 4).

After treatment using cervical stimulation and compression therapy, there was no statistically significant difference between the normal and lymphedematous feet of these 4 children (paired t-test  $P < 0.08$ ) (Table 5).

For these 4 children, a significant reduction in the perimeter of feet was identified on comparing the size of the foot before treatment and after two years of treatment using cervical stimulation and compression therapy (paired t-test  $P < 0.002$ ) (Table 6).

The perimetric differences between feet were reduced or even normalized in 11 children (two-tailed  $P < 0.0001$ ).

## Discussion

The current study reports on a new form of lymphedema treatment in children, evaluated over a 2-year period, which significantly reduced the edema. However, despite this significant reduction, there was still some difference, albeit smaller, between lymphedematous and normal feet. This treatment is based on cervical stimulation with a presumed mechanism of action of stimulating contractions of the lymphangions.<sup>9,10</sup> These stimuli have been used in adults either in association with the manual and mechanical lymph drainage techniques developed by the authors or as the only treatment.<sup>12-16</sup>

Many professionals, including physicians, physiotherapists and occupational therapists, and even the mothers of the patients can be trained to perform this technique. In the first months of life, few difficulties were observed in treating children using stimulation; however, from the age of around six months, the problems increased because the children did not stay still. In this phase, mothers carried out the stimulation while their child was sleeping.

For 10 children, no other forms of treatment, not even manual or mechanical lymph drainage, were used during this period as there was only a small difference between the feet and it was, therefore, decided to only use this simple approach.

**Table 1.** Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet of 10 children before treatment with only cervical stimulation.

Child #	Gender	Position	Lymphedematous (cm)	Normal (cm)	Difference (cm)
1	Male	3 cm	21.5	18	3.5
		6 cm	19.8	16	3.8
2	Male	3 cm	16.8	12	4.8
		6 cm	15.5	13	2.5
3	Male	3 cm	18	14.5	3.5
		6 cm	17.3	13.5	3.8
4	Male	3 cm	18	15	3
		6 cm	16.3	15	1.3
5	Female	3 cm	20.8	18	2.8
		6 cm	17	16	1
6	Male	3 cm	14.5	12	2.5
		6 cm	13	11	2
7	Male	3 cm	13.6	12.5	1.1
		6 cm	12.8	11.8	1
8	Female	3 cm	6	3	3
		6 cm	5	2.5	2.5
9	Male	3 cm	11	9	2
		6 cm	10	8.2	1.8
10	Male	3 cm	17	15	2
		6 cm	16.2	14	2.2

Paired t-test  $P < 0.0001$ .

**Table 2.** Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet of 10 children after two years of using only cervical stimulation.

Child #	Gender	Position	Lymphedematous (cm)	Normal (cm)	Difference (cm)
1	Male	3 cm	19	19	0
		6 cm	17.2	17	0.2
2	Male	3 cm	15	15	0
		6 cm	15.5	16	-0.5
3	Male	3 cm	16	17.5	-1.5
		6 cm	14.3	15	-0.7
4	Male	3 cm	15	15	0
		6 cm	16	16	0
5	Female	3 cm	17	18	-1
		6 cm	16	16	0
6	Male	3 cm	13	13	0
		6 cm	11.8	12.3	-0.5
7	Male	3 cm	14	14	0
		6 cm	12.8	13	-0.2
8	Female	3 cm	10	11	-1
		6 cm	9	9.8	-0.8
9	Male	3 cm	13	13	0
		6 cm	11.5	11.8	-0.3
10	Male	3 cm	17	16.3	0.7
		6 cm	15	15	0

Paired t-test  $P = 0.02$ .

Inelastic compression stockings were used in 4 children who had suffered from the disease for more than three years because these children have greater clinical evidence of fibrosis. A significant reduction in the edema was observed on comparing the lymphedematous foot before and after treatment, and no significant difference was seen comparing the affected foot with the unaffected foot at the end of treatment. This suggests that the association of manual cervical stimulation with inelastic compression therapy has a synergistic effect in treating these children.

One objective of this study was to give effective treatment over a short period of time. However, for this study, treatment over a long period of time was necessary. In some cases, other forms of treatment were required to control the lymphedema because of, for example, absence of family care, infection, or fibrosis. These included manual and mechanical lymph drainage. However, with these measures life-long control of the lymphedema can be achieved. The use of a compression garment is not well accepted by children because they think their peers will tease them about their disease and exclude them from the social group when they see it. When possible, we normally choose the association of manual or mechanical lymph drainage with cervical stimulation,<sup>9,10,13,15</sup> in order to avoid the use of compression garments,<sup>12</sup> even though this latter is the best treatment option.

Great precautions must be taken against infections, in particular, erysipelas which is very common. The mothers are counseled to take their children immediately to a pediatrician if they suspect any type of infection.<sup>17</sup> They are also recommended to bring up their children normally and let them participate in all common childhood activities. However, it is important for them to take special care of wounds to the legs and mycoses. Information on general care is crucial and so these aspects are emphasized at all clinic visits.<sup>18</sup> The objective is to maintain the child's quality of life which is frequently affected in patients with lymphedema.<sup>19</sup>

There is a consensus that an association of therapies should be used in the treatment of lymphedema. However, before starting treatment, it is important to investigate the possibility of other associated conditions so that appropriate measures can be taken.<sup>3,8,20</sup>

This study opens up a new perspective in the treatment of lymphedema in children. The authors suggest that specialized treatment centers for these children should consider evaluating this approach. The independence that this approach offers the family, with the mother being trained to help treat her child, is extremely important.

**Table 3. Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet of ten children before treatment and after two years of treatment using only cervical stimulation.**

Child #	Gender	Position	Difference before treatment (cm)	Difference after treatment (cm)
1	Male	3 cm	3.5	0
		6 cm	3.8	0.2
2	Male	3 cm	4.8	0
		6 cm	2.5	-0.5
3	Male	3 cm	3.5	-1.5
		6 cm	3.8	-0.7
4	Male	3 cm	3	0
		6 cm	1.3	0
5	Female	3 cm	2.8	-1
		6 cm	1	0
6	Male	3 cm	2.5	0
		6 cm	2	-0.5
7	Male	3 cm	1.1	0
		6 cm	1	-0.2
8	Female	3 cm	3	-1
		6 cm	2.5	-0.8
9	Male	3 cm	2	0
		6 cm	1.8	-0.3
10	Male	3 cm	2	0.7
		6 cm	2.2	0

Paired t-test P=0.0001.

**Table 4. Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet of 4 children before using cervical stimulation and compression therapy.**

Child #	Gender	Position	Lymphedematous (cm)	Normal (cm)	Difference (cm)
1	Female	3 cm	18	15	3
		6 cm	16.3	13	3.3
2	Female	3 cm	18	15	3
		6 cm	16.7	14	2.7
3	Male	3 cm	22	20.3	2.3
		6 cm	19.5	18.3	1.2
4	Female	3 cm	18.5	15.5	3
		6 cm	15	14	1

Paired t-test P<0.0002

**Table 5. Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet after two years of treatment using cervical stimulation and compression therapy.**

Child #	Gender	Position	Lymphedematous (cm)	Normal (cm)	Difference (cm)
1	Female	3 cm	18	18	0
		6 cm	16	16.5	-0.5
2	Female	3 cm	17	17.5	-0.5
		6 cm	16	16	0
3	Male	3 cm	21.5	22	-0.5
		6 cm	20	20	0
4	Female	3 cm	15	15.3	-0.3
		6 cm	14.2	14	0.2

Paired t-test P=0.08.

**Table 6.** Perimetric measurements (3 and 6 cm from the base of the big toe along the dorsum of the foot) of normal and lymphedematous feet of 4 children before treatment and after two years of treatment using cervical stimulation and compression therapy.

Child #	Gender	Position	Difference before treatment (cm)	Difference after treatment (cm)
1	Female	3 cm	3	0
		6 cm	3.3	-0.5
2	Female	3 cm	3	-0.5
		6 cm	2.7	0
3	Male	3 cm	2.3	-0.5
		6 cm	1.2	0
4	Female	3 cm	3	-0.3
		6 cm	1	0.2

Paired t-test  $P < 0.002$ .

## Conclusions

Cervical stimulation is a new option in the treatment of primary congenital lymphedema and its association with compression stockings has a synergistic effect in reducing the volume of feet affected by lymphedema.

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